

Anglia Ruskin University

**PERCEPTIONS OF FQOL OF
FAMILIES WHO HAVE A CHILD WITH TOURETTE SYNDROME**

by

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in Partial Fulfillment of the Requirements for the degree of PhD
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ABSTRACT

The Family Quality Of Life (FQOL) of families who have a child with Tourette Syndrome (TS) has not yet been investigated globally. The purpose of this study was to gather data and listen to the voices and opinions of families of children with TS in order to examine the extent to which the existence of a child with TS, in the family, affected their FQOL.

The research was conducted according to the mixed methods approach, combining quantitative and qualitative methods. Data was collected concerning the research population that included all the families who have a child with TS living at home, and registered at the Tourette Syndrome Organisation in Israel. Additionally, semi-structured interviews were conducted with 10 of these 25 families of children with TS. The questionnaire was analysed statistically and the interviews were analysed using a content analysis method.

Conclusions derived from the research findings allowed the emergence of the ACEIST model, including five new sub-domains of FQOL that reflect the experiences of the studied families: Advocacy, cooperation, explanation, information, and severity of TS. This is a new way to view FQOL of families having a child with TS, very precise and operative and specific to those families. This ACEIST model fills the gap in knowledge identified in this field, contributing to knowledge regarding FQOL of families who have a child with TS. The new model has practical implications for the education, social and health policies in Israel regarding families who have a child with TS.

CHAPTER 1: INTRODUCTION

1.1 Preview

Chapter 1 introduces the research topic, giving Justification for the research, research context, explaining the research aims and focus. It begins with the background that led to the research, indicating the reasons for and significance of research examining the Family Quality of Life (FQOL) of families who have a child with Tourette Syndrome (TS) in Israel, within the relevant corpus of knowledge.

The chapter presents the main themes of the research, discusses the research questions, and establishes the theoretical context of the study. The research boundaries are set.

1.2 Justification for the Research

Working as a social worker for ten years in Israel, then as a lecturer of special education at various teachers' colleges in Israel, and more recently as the principal of a high school for the learning disabled, I have often encountered parents of children with various learning difficulties. Some of these families included children who had Tourette Syndrome. These parents reported many difficulties relating to the process of diagnosing the syndrome. They reported many difficulties within the educational system, which was not well-informed and did not understand the needs of these children and their families, and preferred not to cope. In some cases, the educational framework decided to exclude them from mainstream education into special education frameworks. These parents needed to receive information and guidance to effectively treat and cope with the phenomenon. They felt frustrated with society's lack of awareness regarding the phenomenon. Often, they found themselves trying to explain and excuse their children's 'unacceptable' behaviour to friends, teachers, and family members. Some of the families that I have met somehow managed to cope successfully, supported their child with Tourette Syndrome, and to assist with the child in coping with the phenomenon. Other families were unable to adjust and cope, generally leading to high levels of stress in the family, and in certain cases, a break-up of the family.

Three years before I decided to embark on my PhD, I learned about Tourette Syndrome. My decision to undertake research about the families of children with TS came from my holistic point of view that children with any disability, no matter how severe, can be understood, treated and helped through the strengths of their families.

To do so, I needed to undertake research that could provide information about the family's perceptions of their Family Quality of Life (FQOL). This information could assist in constructing intervention programmes to help the child with TS and improve family quality of life.

1.3 Research Context

There is a growing understanding among disability researchers that disability is socially constructed, meaning that it is defined variably across cultures (Olkin, 1999; Ferguson, 2001). Culture is defined as *'everything that people have, think and do as members of society'* (Ferraro, 1995, p. 16). Culture influences the way in which families define and experience disability. Cultural differences and parental perceptions are considered key factors in interactions between parents and professionals. Research and practice regarding child development reveal how important it is to understand the family's context and to recognise the family's cultural backgrounds.

Socio-cultural influences on the parents' perceptions of their child with a disability are recognised within an ecological model (Bronfenbrenner, 1979), a transactional model (Sameroff & Fiese, 2000), and relationship-based approaches (Bertacchi, 1996; Moss & Gotts, 1998). These models consider the child as a point of focus within the immediate context of his/her family, community, and social services. These models also view the family, and their cultural background, as having a critical role in the development of their children with disability (Burnett & Meares, 2000).

Israeli culture is heterogeneous, with a diverse population of immigrants from five continents and their descendants and more than 100 countries, and characterised by significant subcultures such as the Palestinians, the Russians, and the Greek and Armenian Orthodox, each with its own newspapers and cultural networks, (Cohen, 2000).

Official government documents (Jerusalem Institute for Israel Studies, Jerusalem Statistical Yearbook 2009/10) show that the population in Israel in 2009 was approximately 7,503,800. 75.4% of the population were Jewish, 20.8% were Arabs. Hebrew and Arabic are the nation's official languages.

Health care in Israel is administered by a small number of organisations with funding from the government. All Israeli citizens are entitled to the same benefits, regardless of which organization they are a member of, and treatment under this package is funded for all citizens regardless of their

financial means. Since 1973, all employers in Israel participate in the medical insurance of their workers, by means of a direct payment to the Health Maintenance Fund in which the workers are members (Surviving Israeli Healthcare , 2009)

Education in Israel is mandatory from age five to fifteen. The state runs both religious and secular schools; 70 per cent of children attend secular schools. A separate education system provides education for Arab children, where the language of instruction is Arabic (Cohen, 2000). Israeli schools today may be classified in general either as state schools, (schools that emphasis focus and rigour in study. Many state secular schools specifically design their programs to prepare students for '*Bagrut*' Matriculation examinations for university) or as state-religious schools (have similar curricula to state secular schools, but they supplement education with religious Torah studies) or they may be part of the separate educational structure maintained for Arab students, though some Arabs study in the Jewish system. Arab schools (including Christian, Druze and Islamic schools) teach students in Arabic and supplement Israeli education with Arabic history. Arab schools are known to have strict rules, and instructors have absolute authority in the classroom. Although the Ministry of Education tries to provide high quality education for all students, Arab schools receive fewer government funds than other schools (Cohen, 2000). The state and state-religious schools are under the supervision of the Ministry of Education Culture and Sport. The language of instruction in Jewish Schools is Hebrew and in the Arab schools is Arabic. Higher education is supervised by the Council for Higher Education. The Council has the power to recognise institutions of higher education and to grant authority to award degrees.

In Arab schools all students begin to study Hebrew in Year 3, at the age of 9. A second foreign language such as English or French may be added in the Year 4. Hebrew is the language of instruction in all Jewish schools, where Arabic or French are taught from Year 7. Religious studies are required in all types of school. Teaching methods are determined by school principals, teachers, and local authorities, but the content of the overall curriculum is strictly laid down by the Ministry of Education (Cohen, 2000).

Women in Israel are well represented in many fields, both traditional (teaching, nursing, child care), and non-traditional (law, politics, the military). Today, Israeli women are more likely to be found in child care facilities.

The most common family unit consists of a nuclear family. In more traditional families, grandparents are sometimes included within this unit. The extended family plays an important role in helping to raise the children, but the mother generally takes primary responsibility (Cohen, 2000).

Within this socio-cultural context, this research sought to examine how families of a variety of cultural contexts, with children who have TS perceive their family quality of life. Ultimately, only Jewish families agreed to participate in this research.

1.4 Aims and Focus

1.4.1 Introduction

Having a child with Tourette (also called Tourette's) Syndrome (TS) seriously challenges parents who aspire to raise a 'normal' family, because of the stresses and strains engendered by the unusual behaviour of the child with TS (Greene, 1996). Children with TS may experience a wide range of psychological, behavioural and psychiatric problems in addition to vocal and motor tics. These difficulties also lead to interpersonal communication problems (Carter et al., 2000), feelings of anxiety and mood-changes (Robertson, 2002), and may be accompanied by difficulty in concentrating, hyperactivity and obsessive behaviours (ibid). These children usually find it hard to adapt their behaviour to the environment and their particular characteristics may have negative impact on the child's functioning at school, since they create difficulties with self-regulation and in social integration and acquisition of education (Greene, 1996). In addition to their difficulties in daily functioning in school, (Piacentini et al., 2003) children with TS exhibit significant problems with labile emotions, impulsivity, and aggression directed at others. Aggression may be expressed through temper fits that include screaming, punching holes in walls, threatening others, hitting, biting, and kicking (Carter et al., 2000; Swain et al., 2007). Often these phenomena create much consternation to teachers and anguish both for those with TS themselves and to their families (Carter et al., 2000).

Usually this situation engenders confusion, helplessness and sometimes an inability to maintain 'normal' family life. Instead of turning to counselling or seeking help, some families withdraw within themselves (Tourette Syndrome Organisation, Israel, TSOI, 2008). Research conducted among families of children with disabilities in Israel found that most of the families who have children with disabilities receive no help dealing with their child (Turnbull et al., 2004). Thus, raising a child with TS has significant implications for the family's everyday life.

From my experience as a social worker for ten years in Israel, then as a lecturer on special education at various teacher-training colleges, and more recently as the principal of a high school for pupils with learning disabilities, I have often encountered parents of children with various learning difficulties. Some of these children were diagnosed as having TS. Their parents reported obstacles they encountered in the process of diagnosing the syndrome. They also indicated that they had many difficulties within the educational system, which was not well-informed concerning the syndrome and was not fully aware of the needs of these children and their families, preferring not to cope with the challenge. In some cases, the education system decided to reject and exclude children with TS. These parents needed information and guidance regarding ways to treat and cope with the phenomenon. They felt frustrated with society's lack of awareness and responsiveness regarding the phenomenon. Often, they found themselves trying to explain and excuse their children's 'unacceptable' behaviour to friends, teachers, and family members.

Surprisingly, there were families that I met who nevertheless appeared to cope successfully, supporting their children with TS, and managing to assist them in coping with the phenomenon. Other families who were apparently unable to adjust and cope, developed high levels of stress, and in certain cases, this led to the break-up of the family (TS Organisation Israel, TSOI, 2008). In this sense, living with a child with TS challenges family life. Additionally the integration of a child with TS in school challenges the various professional personnel within the educational system. The apparent inability of schools in Israel to include children with TS calls for an examination of appropriate ways to accept and deal with these children and their families.

1.4.2 A gap in knowledge

Much research and theoretical literature relates widely to TS (Dreher, 1996; Abelson et al., 2005; Swain et al., 2007), including consideration of the ways to diagnose children having TS (Dreher, 1996; Pauls et al., 1993; Merck Manual of Medical Information, 1997), and treat them (ibid).

More precisely, the literature provides ample knowledge regarding the medical (ibid) as well as the social aspects (Sukhodolsky et al., 2003; Bawden et al., 1998; Carter et al., 2000) of children with TS. Additionally, there is some knowledge in the literature about Quality of Life (QOL) of children with TS (Browne et al., 1997; Turnbull et al., 2000; Elstner et al., 2001; Goodman et al., 2006), but as far as the researcher could verify there is an apparent dearth of literature concerning FQOL of families with children who have TS. This suggests that there is a gap in knowledge regarding FQOL relating to families with children with TS. This research seeks to fill this gap in knowledge by investigating perceptions of FQOL in families who have a child with TS. The researcher's professional experience suggested that the lack of awareness regarding the phenomenon bears potential social and functional risks.

These risks reside within the family environment, among extended family members, friends, and various professionals that further encumbers the family's ability to function on a daily basis. This may cause the nuclear family members to distance themselves from places that are full of people, preferring to remain at home, isolated. The research may therefore provide and provoke additional awareness of this phenomenon, and the need for greater knowledge and understanding.

The research aimed to provide evidence based knowledge which may offer guidance to families and educational institutions concerned with the treatment of these children and their families.

Accordingly, the thesis aimed to address two main questions:

1. Which FQOL domains are perceived by family members to be affected by the fact that they have a child with TS?
2. How do different families raising a child with TS perceive their FQOL?

1.4.3 The rationale for the research

As noted above, the study attempted to fill an identified gap in knowledge concerning the FQOL of families who have a child with TS. TS is defined in the research literature as an inherited (genetic), neurological disorder characterised by repeated involuntary movements and uncontrollable vocal (phonic) sounds called 'tics'. Symptoms generally appear between the ages of five and seventeen years old (Stern et al., 2005; Albin & Mink, 2006). Internationally, studies have indicated that TS occurs in around 1% of children in mainstream schools (Robertson, 2002; Stern et al., 2005).

According to the official site for TS in Israel (TSOI ,2008), only 500 families (Apter et al.,1992) are registered on a voluntary basis as having children with TS, out of a known population of 3,200 persons diagnosed with TS in Israel. The voluntary registration of a family may imply a degree of acceptance of the fact. However, when families do not register, this might imply that they are embarrassed, ashamed, hiding the phenomenon, and/ or find it difficult to cope with the affected child

Families of children with TS are usually in a state of imbalance (Cohen et al., 1988; Wilkinson et al., 2001) mainly because they have to live with the specific disability that is expressed not only in vocal and motor tics but also often by conduct disorders, such as Obsessive Compulsive Disorder (OCD), Attention Deficit and Hyperactive Disorder (ADHD) or Attention Deficit Disorder (ADD), Learning Disabilities (LD), insomnia, depression, low self-image, aggressive behaviour, and various phobias (Greene, 1996; Stern et al., 2005).

Yet the family's difficulties do not focus solely on the perceptible phenomena of the child with TS, which can be embarrassing to the child and the family. In most cases, the family must also deal with an educational system that does not know how to contain this phenomenon and is consequently unprepared to absorb and educate children with TS. This means that schools in Israel are often unwilling to accept children with TS, and families are unable to find an appropriate framework for their care and education.

The Israeli Ministry of Education has difficulty defining the phenomenon, and these children fall between the cracks. They are not specifically included in the Ministry Of Education, General Manager's Circular (2004): Populations with Special Needs' as a population with special needs. Children with TS have an IQ within the 'normal' range, but the complexity of their verbal and motor tics, in addition to other academic and behavioural problems, leads the educational system to 'give up' and even ignore the problem. In Israel, it seems that the system expects the family to cope instead of designing intervention programmes which would help to adapt the system to the needs of the child with TS and the family. The families, who have a child with TS, are left alone, with no appropriate catering for their needs. This fact, augments the difficulty of coping within the family with the outbursts of anger that these children exhibit, their movements, sounds and restlessness, and may lead the family to instability and defective functioning. The daily difficulty involved in containing the child, with TS, at home, in the educational framework, and in various social frameworks, can easily cause a feeling of frustration among family members, which consequently may affect the entire family as a unit.

In an attempt to explore these issues in the context of the Israeli family, the researcher employed an inductive approach to attain two central aims:

1. To understand how the FQOL of families who have a child with TS is affected by this syndrome.
2. To fill the gap in knowledge regarding perceptions in families with children with TS concerning their FQOL.

A theoretical foundation was established that would support and guide the research process. This foundation was derived from:

1. Behavioural psychology
2. Systems theory
3. The ecological model

It was hoped that the research could contribute to knowledge by offering new insights and new thinking strategies that would inform and facilitate the design of appropriate intervention programmes to improve the FQOL of families, who have a child with TS.

The present research therefore examines perceptions of FQOL in families of children with TS in Israel, indicating the domains of FQOL which might be affected by raising a child with TS in the family, with the purpose of indicating directions for future research and therapeutic efforts to help these families improve their FQOL and the successful integration of their children within schools and society.

1.4.4 Methodology and research boundaries

The research was conducted in Israel, in the years 2006-2007. Since the research sought to examine the FQOL of families who have a child with TS, and with no pre-determined hypotheses available, an inductive research design approach was adopted (Creswell, 2003).

A mixed-methods research strategy was implemented to provide a holistic view of this phenomenon (ibid; Ortiz & Greene, 2007; Curry et al., 2009). The research was conducted in two stages. Stage 1 applied a quantitative approach employing a closed-ended questionnaire, to measure the extent to which families with a child with TS were satisfied with their FQOL in the five FQOL domains (Turnbull, 2004). In Stage 2, a qualitative approach, (Creswell, 2003; Ortiz & Greene, 2007) was used to address related issues and deepen the understanding of the families perceptions of their FQOL. Interviews (Oppenheim, 1992; Scheurich, 1995) were conducted with the families to ascertain their views, opinions, and feelings. Statistical analysis (Murphy & Mayers, 1998) was employed to assess the data collected through the closed-ended questionnaire, while content analysis (Carley, 1990) was employed to analyse the opinions and views expressed in the interviews. The quantitative part of the research had limited value. In contrast, the qualitative part of the research was considered more significant.

The research population, families of children with TS over age seven were chosen for this research for a number of reasons. Several studies indicate (Dreher, 1996; Robertson & Baron-Cohen, 1995; Abelson et al., 2005; Swain et al., 2007), that families of children with TS begin experiencing difficulties after the child reaches age six-seven because it is only at this age that the verbal and motor tics begin to appear (Dreher, 1996; Abelson et al., 2005; Swain et al., 2007) and TS is usually not diagnosed until this age (Robertson, 2002). As children begin schooling in Israel by the age of six/seven, then, until this time, families are generally not aware of the full implications of the child's TS. The family may go through a sudden change, with unclear ramifications regarding the family's future functioning, which entails feelings of frustration, helplessness and fear.

A second consideration in the choice of the research population is that in addition to the motor and vocal tics, children with TS may also present the following symptoms: Attention Deficit Hyperactive Disorder (ADHD), or Attention Deficit Disorder (ADD), (Robertson, 2002), Obsessive Compulsive Disorder (OCD) (ibid), and other difficulties (Dreher, 1996; Albin & Mink, 2006). Consequently, the research needed to take into account these and other similar conditions. A lack of specific intervention programmes for children with TS at school has made it difficult for parents to ensure the inclusion of their children with TS within the education system. It was hoped that the research might therefore provide valuable information for educators in this field to enable them to cope more successfully with these children.

1.5 The Structure of the Thesis

The thesis is divided into 7 Chapters as follows:

Chapter 1: The present chapter introduced the reader to the research topic, and the gap in knowledge regarding this topic. It outlined the research, and the characteristics of the research population. It explained the questions which the research attempted to answer, and indicated the methodology that would be used to gather and analyse relevant data.

Chapter 2: This chapter describes five areas of knowledge chosen for their relevance to the research topic, as they are considered in theoretical and research literature. The five areas of knowledge relate to: Tourette Syndrome (TS) [including Parenting a Child with TS, Children and Adolescents with TS, The Child with TS at School, Siblings of Children with TS]; Family Quality of Life (FQOL) [including consideration of the Modern Family, The Family in Israel and the definition of FQOL and its domains] and Systems Theory [Including Family Systems Theory] and the Ecological Model. This combined corpus of knowledge provided the foundation for the construction of the research's conceptual framework.

Chapter 3: describes the construction of the research's conceptual framework and its constituent elements concluding with a graphic representation of this framework.

Chapter 4: explains the choice of research methodology and the manner in which it was employed to collect and analyse the research data, including a description of the research methods and tools. It relates to issues of reliability and validity and whether or not it is possible to generalise from the research results to other similar situations. Details are given regarding the research population and the way in which the research dealt with ethical issues involved in conducting the research.

Chapter 5: presents the research findings and their analysis in relation to the sequence of research questions and according to the different tools used to collect the data.

Chapter 6: discusses and interprets the data presented in Chapter 5 indicating the relationship of these data to relevant theories and research in these fields.

Chapter 7: draws factual and conceptual conclusions from the discussion in Chapter 6 and employs these conclusions to formulate a new model outlining the characteristic elements of FQOL for families who have a child with TS as they emerged from this research. It suggests operative recommendations as a result of these conclusions and indicates possible directions for future research. Finally it summarises the contribution of the present research to extant knowledge.

The thesis concludes with an Epilogue, describing my personal journey as I pursued the research.

1.6 Chapter Summary

This chapter identified a gap in knowledge concerning the families of children with TS. It provided the rationale for the study and introduced the research aims and focus, the context of the study and its methodology. The next chapter discusses the main theories that underpinned this research.

CHAPTER 2: THEORETICAL PERSPECTIVES

2.1 Introduction

This thesis explores the concept of FQOL and how it is affected by the existence in the family of a child who has TS.

Accordingly this chapter suggests a broad, though not exhaustive, spectrum of the definitions and explanations of TS. Additionally it discusses definitions of FQOL and reviews literature relating to the five family domains of FQOL mentioned in the Introduction (family interaction, parenting, emotional well-being, physical/material well-being, and support for the family member with a disability), which provided the main focus for the research data analysis. It also clarifies the concept of family systems, which formed an essential building block for the establishment of the methodological foundation for this research and explains Systems Theory, which is the theory used in this thesis to describe FQOL.

Thus, the chapter captures, depicts, discusses, and analyses the different theoretical views and research experience in the relevant fields of knowledge, engaging with current ideas, issues and trends relating to the research topic. The concluding section introduces the conceptual framework of this research based on conclusions from this review.

2.2 Disability in the Family Context in general and in Israel in particular

In some cases, having a child with a disability creates negative family outcomes including added stress and parental depression (Baxter, Cummins, & Yiolitis, 2000; Hayden & Goldman, 1996). Studies have noted poorer family outcomes associated with child behaviour problems. A child's challenging behaviour has been associated with increased burden and stress on families (Heller, Hsieh, & Rowitz, 1997; Simmerman et al., 2001; Saloviita et al., 2003). Maladaptive behaviour and care needs may be more important risk factors for family impact and stress.

Apart from behavioural problems, other types of care demands can create stress and burden for families. Personal care needs (Neely-Barnes & Marcenko, 2004; Plant & Sanders, 2007), adaptive behaviour deficits (Saloviita et al., 2003; McCarthy et al., 2006) and medical needs (Neely-Barnes & Marcenko, 2008) have been associated with negative family impact, stress, and burden.

In addition, the way the family functions in response to the child's disability has a critical impact on family outcomes. Researchers have noted that marital and family functioning may be far more important predictors of parenting stress and depression than the presence or absence of childhood disability. Abbeduto et al. (2004) found that diagnosis-specific differences in parenting stress disappeared when child behaviour, parental coping style, maternal education, family income, age of the child, and number of children in the family were controlled. Kersh et al. (2006) found when controlling for marital quality, neither child functioning nor child behaviours were significant predictors of maternal and paternal depressive symptoms or parenting stress. Smith, Oliver & Innocenti (2001) noted that although poor social skills in the child were a predictor of parental stress, family functioning was a much stronger predictor. These studies point to the need for practitioners to pay attention to parental relationships and overall family functioning because these can be important risk factors for parental stress.

Although several studies have examined the negative impact of poor family functioning, most families of children with disabilities do not experience poor functioning (Lustig, 1997). Families of children with a disability may experience no more difficulties with functioning than other families. Thus, practitioners should not equate poor family functioning with childhood disability. While poor family functioning may be a risk factor for stress when it is present, many families function quite well (ibid).

In Israel families of children with disabilities experience strong feelings and emotional stress (Levy-Shiff, 1997, 1999). Mothers, report continual stress that is a main attribute of such families. It was found that the parent of the child with disability in Israel must be continually aware of the child's situation and possess substantial knowledge regarding the appropriate daily care for the child, particularly in times of distress. Parents need to participate maximally in routine caring, including visits to clinics and also often hospitalisation. All these tasks lead to parental stress (Duvdevani et al., 1997).

In this sense, there is a scholarly consensus that families caring for a child with a disability face many different difficulties especially when the child with a disability has behaviour problems.

2.3 Conceptions of Tourette Syndrome (TS)

TS is a neuropsychiatric disorder named after Dr. Georges Gilles de la Tourette (1857-1904), a pioneering French neurologist, although the first descriptions of motor and vocal tics were reported 60 years earlier in 1825 by the French neurologist Jean-Marc Itard (1775-1838). His patient, the Marquise de Dampierre, was a French noble woman who developed motor tics at the age of seven, and shortly thereafter began to manifest involuntary vocalisations consisting of screams and strange cries. Several years later she developed coprolalia (uncontrolled utterance of inappropriate words and phrases). With this list of problems, namely, motor tics, screams and coprolalia, the Marquise was forced to live in seclusion and continued her involuntary cursing until her death at the age of 86. Some 60 years after Itard's report, Gilles de la Tourette produced a detailed account of several patients with a similar condition, including the Marquise in her later years, which clearly established the existence of this phenomenon. Jean Martin Charcot, one of the leading European neurologists of the 19th century, and Gilles de la Tourette's supervisor at the Salpêtrière Hospital in Paris, adopted his pupil's name for this syndrome (Comings, 1996).

TS is an inherited, neurological disorder characterised by repeated involuntary movements and uncontrollable vocal (phonic) sounds called tics. In some cases, such tics can include inappropriate words and phrases, (Dreher, 1996).

As noted above, the symptoms of TS generally appear before the individual is 18 years old and usually at the age of six to seven years old (ibid; Abelson et al., 2005; Swain et al., 2007). TS can affect people of all ethnic groups, and it appears to be familial (i.e., it 'runs' in families). Recent information appears to indicate that there is likely to be a significant genetic factor responsible for its genesis. Research by Abelson et al., (2005) reviewed some of the genetic research and identified a new candidate gene. Gender is also linked to the expression of TS: boys are significantly more likely to develop TS than are girls; males are affected three to four times more than females (Dreher, 1996).

Male offspring of people with TS have almost a 50% chance of having TS symptoms, and a .99 (almost certain) probability of having either TS, a chronic motor tic disorder or OCD (Pauls et al., 1993). Female offspring had lower probabilities of showing symptoms of these disorders, but there was still a fairly high probability (.70) that a female child of a parent with TS would have one of these disorders (ibid).

Although TS symptoms range from very mild to quite severe, the majority of cases fall in the mild category (Abelson et al., 2005; Swain et al., 2007). According to the Merck Manual of Medical Information (1997), the first symptoms of TS are usually facial tics, commonly eye blinking. However, facial tics can also include nose twitching or grimaces. With time, other motor tics may appear such as head jerking, neck stretching, foot stamping, or body twisting and bending. Those with TS may utter strange and unacceptable sounds, words, or phrases. It is not uncommon for a person with TS to continuously clear his or her throat, cough, sniff, grunt, yelp, bark, or shout.

Additionally, people with TS may involuntarily shout obscenities (coprolalia), or constantly repeat the words of other people (echolalia). They may also touch other people excessively or repeat actions obsessively and unnecessarily (Abelson et al., 2005; Swain et al., 2007). A few of those with severe TS demonstrate self-harming behaviours such as lip and cheek biting and head banging against hard objects. However, these behaviours are extremely rare. Tics alternately increase and decrease in severity, and periodically change in number, frequency, type and location (Bagheri et al., 1999). Symptoms may subside for weeks or months at a time and later recur.

Furthermore, there are two categories of tics: simple tics and complex tics. Simple tics are sudden, brief movements that involve a limited number of muscle groups. They occur in a single or isolated fashion and are often repetitive. Some of the more common examples of simple tics include eye blinking, shoulder shrugging, and facial grimacing, head jerking, yelping, and sniffing.

Complex tics are distinct, coordinated patterns of successive movements involving several muscle groups. Complex tics might include jumping, smelling objects, touching the nose, touching other people, self-harming behaviours, echolalia and coprolalia, which include involuntary outbursts of obscene words or socially inappropriate and derogatory remarks (Dreher, 1996; Kushner, 1999; Cohen & Levi-Pearl, 2006). For those with coprolalia who are trying to deal with society in places such as school, work, or home, just getting through the day can be excruciatingly difficult. People with TS can sometimes suppress their tics for a short time, but eventually tension mounts to the point where the tic escapes. Tics worsen in stressful situations; however, they improve when the person is relaxed or absorbed in an activity. In most cases, tics decrease while the person sleeps (Dreher, 1996; Kushner, 1999; Cohen & Levi-Pearl, 2006).

Most people with TS experience additional problems such as OCD, where the persons feel that something must be done repeatedly, such as hand washing or checking that a door is locked and have obsessively repeated thoughts (Robertson & Baron-Cohen, 1995). Additional problems may include ADHD or ADD, (Robertson et al., 1988; Robertson, 2002), LD which include reading, writing, arithmetic, and perceptual difficulties; problems with impulse control, which can result in overly aggressive behaviours and socially inappropriate acts; or sleep disorders, which include frequent awakenings or talking in one's sleep (Dreher, 1996; Albin & Mink, 2006). In sum, the literature depicts TS as a severe disorder, including symptoms that may have implications on parenting such a child. The, next section delineates the issues involved in parenting children with TS.

2.4 Parents of Children with Disabilities

Since this research focuses on a study of FQOL of families, who have a child with TS, the theoretical background for this examination is supplemented by discussing the issues involved in parenting children with TS. First, the parallels between parenting a child with TS and parenting children with Special Education Needs and/or disabilities will be discussed.

Parents' perceptions on the nature of a disability are determined to some degree, in line with their cultural values. This is also true for their views about the nature of a disability. Common perceptions include envisaging that their children as developing normally, identifying their children's condition as a temporary or passing condition, and perceiving a disability as God's punishment or, conversely, as God's special gift (Reiter, Mar'i, & Rosenberg, 1986; Ryan & Smith, 1989; Mardiros, 1989; Stahl, 1991; Steinberg & Davila, 1997; McCallion & Janicki, 1997; Garcia, Perez, & Ortiz, 2000).

Mexican American parents of children with disabilities believe their child's development to be normal (Mardiros, 1989; Garcia, Perez, & Ortiz, 2000). Some parents defined their child as having a disability and yet developing normally. Mexican American mothers of children with language disabilities were not concerned about their children's language difficulties (Garcia, Perez, and Ortiz, 2000). Although they accepted that their children were developing at a different rate, they had expectations that their children would have better communication skills after age three and the mothers believed that as their children grew older, they would 'catch up' with their peers (Garcia, Perez, and Ortiz, 2000). On the other hand, Mexican American parents of children with disabilities clearly distinguished between being disabled and being normal. Believing that each child with a disability was unique, they perceived their child's condition as a disability, and not an illness or disease (Mardiros, 1989).

Parents of Chinese American children with an intellectual disability had a tendency to see the disability as a temporary problem (Ryan & Smith, 1989). Druze parents in Arab communities, who believe in reincarnation and life after death, also consider the disability to be a temporary or passing condition. Oriental Jewish mothers were greatly confident in their beliefs that a change would happen suddenly and their child would become 'normal' (Stahl, 1991, p. 365). God is perceived by some parents, especially those coming from strongly traditional cultural groups, as the agent of disabled children. Hispanic parents view disability as a punishment for wrongdoing (Mardiros, 1991; Steinberg & Davila, 1997). Oriental Jewish mothers of children with disability believed that meeting a misshapen person or unclean animal during pregnancy might affect the development of the unborn child (Stahl, 1991). Parents from Mexican American, Jewish and Arab communities, may consider themselves or their families as the agent that caused the disability.

Mothers of children with intellectual disability experience more depression than those of children who develop normally (Olsson & Hwang, 2001). When the parental experience was examined across diagnoses, differences were noted. Parents of children with Down syndrome have been found to experience less stress (Ricci & Hodapp, 2003), depression (Abbeduto et al., 2004), and pessimism (Lewis, *et. al.*, 2006) than parents of children with other diagnoses, particularly autism. Parents of children with disabilities were found to have poorer parenting efficacy (Kersh et al, 2006), poorer parental mental health (Herring, et al., 2006), and poorer marital adjustment (Simmerman *et al.*, 2001). It seems that since Down syndrome can be diagnosed either during pregnancy or at birth, rather than at an older age – as is the case of TS, which is diagnosed at the age of 6-7; this can explain the differences in parents' reactions to their child's disability.

2.5 Parenting a Child with TS

With regard to the coping abilities of families of children with TS in Israel, few studies have been conducted regarding the impact of the birth of a child with TS on the family life pattern (Duvdevani et al., 1997).

Two central aspects have been investigated with regard to processes that occur in the family of a child diagnosed with TS: 1) the various stages of the parents' responses; 2) consequences for siblings of the child. The model offered by Wanlass and Fortier (1984, in Duvdevani et al., 1997), which describes the passage of the family of the child with disability through a number of stages from the time of discovery of the disability, reflects similar stages noticed in families of a child with TS, but with a unique emphasis on the syndrome itself and its attributes. The stages indicated are: 'clash', 'denial' and 'sadness', 'focus on the external world' and eventually 'acceptance'.

According to this multi-stage model, the *Clash Stage* is characterised by shock, mourning and fear. Haerle (1992), (who herself developed a seven-stage model of coping with TS) argues that the initial response of many parents to a diagnosis of TS in their children is 'shock'. This response is very common, particularly in cases in which the syndrome appears suddenly, or when parents had not perceived their child's disability as so severe. Nevertheless, even when parents did sense that something was not right for many years previously, hearing the words 'TS' often causes feelings of shock. TS, unlike many other chronic disorders, is a syndrome that is not well known to many people and therefore most parents have difficulty understanding the nature of their child's disorder. A lack of sensation which often arises as a result of discovering that the child has TS, may help parents avoid being swallowed up by feelings of distress and sadness (Robertson et al., 1988). It also apparently prevents parents from accepting the full meaning of the syndrome and its attributes. At this stage, parents have difficulty absorbing new information provided to them, and therefore during this stage, many neurologists tend to transmit test results and information in a short and simple fashion (Robertson et al., 1988).

In contrast to these responses of shock, sometimes, as a result of the unique nature of development of the syndrome, the initial response of parents to discovering their children's condition is a sense of profound 'relief' (Shimberg, 1995). Because of the unique characteristics of the syndrome, parents have often been aware that something was not right with their child, for many years before diagnosis (Robertson et al., 1988). Since, in most cases, TS is not diagnosed until the child is seven to eight years old; this means that parents have often already become concerned about the child's early development before they approach a neurologist (Shimberg, 1995), they become worried that something is not right and consequently the eventual diagnosis of TS may be less menacing than their most terrible imaginings (ibid).

As mentioned previously, TS is often accompanied by other disorders such as ADHD, which generally appear two and a half years before the tics. This complex of disorders may lead parent to feel that their child is exceptional, without knowing the source of the abnormality, (Greene, 1996). Since children with TS have a 'normal' appearance, people in the environment who are not familiar with the syndrome tend to assume that the child is behaving this way on purpose. Also, the environment often tends to assume that the parents are at fault for their child's problem, and that they have raised the child inappropriately. Therefore, when a name is given to the behavioural patterns of their children, this helps parents understand that their child is not 'crazy', that they are not at fault for their child's situation, that the child's syndrome has a scientific name, and that there is something that can be done to help the child, such as pharmaceutical treatment. These insights often lead to a profound sense of relief (ibid).

Nevertheless, there are parents who do not feel any relief, but rather feel shock and even a sense of mourning because they know that their child has TS. Similar to other families who show signs of 'mourning' and 'loss' when they learn that their child is disabled, the parents of a child with TS also need time to express grief for their child: the fact their child suffers from tics and will always be different from other children around him (Shimberg, 1995). They also mourn for the other children they had previously

considered having, and are afraid to have because of the condition, and they mourn for themselves (ibid), with a deep sense of loss of the 'perfect child', and the 'wonderful family life', that they could have had. The parent mourns the years ahead, anticipating struggles and pressure. Nevertheless, these feelings usually decline gradually when the parents' fantasy that their child will be perfect is exchanged with a more realistic fantasy (ibid).

At the *Denial Stage* - one of the attributes of the syndrome is the appearance of repetitive episodes involving many repeated gestures. This attribute of TS may cause parents, at this stage, to believe that the tics are a 'bad habit', in an attempt to deny the diagnosis provided by the doctor (Haerle, 1992). When tics disappear for periods of time, the 'denial' mechanism is reinforced, and parents feel that their child does not have TS. As a result, sometimes weeks or even months can be wasted when parents continue to deny the diagnosis and their child is prevented from receiving physiological treatment and emotional support, which could help the child himself and the entire family (ibid). The length of the denial period varies from family to family, and the therapist should respect the family's feelings and allow it time to grieve and deny the reality until they reach the stage of acceptance. It is important to provide the family with reliable information about the disability and to allow them to understand and accept it at their own pace.

There is actually also a positive facet to the denial mechanism. Denial helps parents repress their feelings of anxiety and stress. In an extreme case, parents who tend to deny their children's syndrome will continue to act as if nothing has happened (ibid). However, most parents tend to just attribute less importance to the problem and to fantasise that their children will 'get better' naturally (ibid). Gradually, they learn more and more and begin to accept the syndrome and the need for the child and the family to receive assistance.

The Sadness Stage - this stage usually involves anger, guilt and helplessness. After the feelings of shock and denial subside, many parents feel that they are overwhelmed, overburdened and helpless in face of the disorder. Since TS is a complex syndrome, and often appears with other comorbid syndromes, which create additional difficulties for the child, parents tend to feel that they do not know how to help their child or where they should begin (Haerle, 1992). Many parents tend to feel very impotent when they think of what lies ahead, namely, continual problems in school and society for the child, endless meetings with doctors and neurologists, and the side effects of medication (ibid).

Along with the feeling of helplessness, often this stage is accompanied by feelings of 'guilt'. Many families coping with a child with a disability feel guilty, and this is true too for families with children with TS. At this stage, their perception and understanding of the causes of the syndrome may engender guilt feelings (Pezerstone, 1987). For example, since one of the factors is genetic, parents tend to begin to study their family history trying to clarify if one of their family members had TS. As a result, parents may blame themselves or their spouse for not knowing that the syndrome was hereditary or for the lack of knowledge that a family member had TS. Guilt feelings are worse when one of the parents has TS (ibid).

Parents may also feel guilty because of their own responses. Often parents criticise themselves for the way they treated their children before the diagnosis. Since the tics often begin a long time before diagnosis of the disorder, many parents may have punished their child for their tics and their other forms of apparently uncontrollable behaviour. Guilt may even be felt for the frustration and anger that they felt towards their children because of their strange behaviour, and because often they did not respond or intervene when other people or children taunted their children because of the tics (Haerle, 1992).

Parents might also blame the quality of their parenting. Because of the perceptions of doctors and psychologists who used to indicate that the parents' approaches and behaviour may be the source of their child's disorder, many parents were made to feel guilty (Pezerstone, 1987). Many parents of TS children recount that even after the diagnosis of their child's neurological disorder; they still blame themselves for being 'bad' parents.

At this stage, another common feeling among families of children with disabilities is 'anger'. Anger that arises among parents of children with disabilities includes anger towards others, towards professionals and even towards the child himself. Parents of a child with TS tend to feel enraged, particularly towards various professionals, with whom they met before their child was diagnosed, and who wrongly diagnosed their child's problem or did not attribute importance to the disorder (ibid).

These feelings are often accompanied by anger at the environment because of the taunting that the child endures and that the entire family tends to absorb (Shimberg, 1995). This feeling may lead a parent to cut off communication with the entire environment, including the school staff, doctors and others who could support the child and the family, and may, consequently, lead to isolation and depression (Shimberg, 1995).

Another typical response at this stage among parents of children with TS is 'holding a grudge'. Many parents describe increased feelings of jealousy and injustice (Robertson & Baron-Cohen, 1995) as they compare their child and family to other families in which the children are 'normal'. For example, many parents tend to be jealous of families with 'normal' children who are able to go out into society and immediately fit in, whereas families of children with TS often encounter stares from the environment in response to the child's tics, (ibid). Parents of a 'normal' child may not always feel a need to engage with the school staff, whereas parents of a child with TS must encounter many difficulties with the staff regarding their child's adjustment to school and the classroom environment. Other children generally assimilate socially and have a large number of friends, while children with TS tend to have various social difficulties.

As a result of all of the above, the life of a family with a child with TS is more difficult and may lead parents to develop hostility, which could even be directed towards the child himself, for making their life become so difficult. These feelings are 'normal', but nevertheless, they may lead to the parent becoming distant from the child, to feelings of self-pity, and to distancing from society, which as noted decreases the possibilities to receive support (ibid).

The stage of *Focus on the External World* is typified by searching for information, making choices and weighing options, planning and formulating plans, accepting reality, and a sense of relief. The *Acceptance* stage is typified by a feeling of family solidarity and recognition of the special needs of the child with TS (Haerle, 1992).

In these last two stages, parents of a child with TS tend to begin to adjust to the situation and begin to create a realistic picture of the child's problems, and also of the child's strengths, focusing on practical methods of dealing with the situation. At this stage, most parents try to get as much information as possible regarding TS by reading books and joining various organisations. These stages are essential in the case of TS, since the syndrome has not been correctly understood for many years, and there is much erroneous information, misconceptions and prejudices that circulate concerning the disorder and its traits (ibid).

Since the syndrome is so complex, parents need to receive updated information regarding the disorder in order to know how to live with it in the future. At this stage, the emotional storm abates, and parents are more able to accept and absorb information regarding TS (Haerle, 1992).

As mentioned before, these later stages are essential, since they seriously alleviate the prior feelings discussed above, such as guilt and anger. Receiving information helps the family of the child with TS to learn about the child's life, and prepares the parents mentally for changes that the family will be experiencing. Furthermore, by learning and receiving information about TS, the parents can help their children better understand the disorder and can help them live with the disorder and its symptoms (ibid).

Different parents respond differently during the various stages. Hence, the feelings that are experienced by parents of a child with TS during the various stages are experienced to different degrees and in different ways in different families. One of the factors affecting the way that a family responds is the severity of the disorder, expressed in the severity of the motor and vocal tics, and the severity of the comorbid disorders. The higher the degree of severity, the higher will be the level of anxiety and negative (Leckman et al., 1998). An additional factor which affects the nature of the family's response is the degree of psychological flexibility of each of the parents, and their ability to return to their previous level of functioning, along with the ability to accept support from the extended family, friends, and professionals, who can help the parents and the entire family deal with their emotional responses at the various stages, in a better and more effective manner (ibid).

For many families, learning that the child has a condition or 'medical problem' is both a relief and a source of fear, grief, and guilt (Leslie, 2005). Relief is created since the disorder is recognised and has a name and an explanation is provided for the strange behaviour of their child, and it becomes clear that the child's misbehaviour is not a reflection on their parenting skills. Fear is experienced for their child's future and grief over the loss of the anticipated 'perfect' child (ibid).

In addition to Haerle's (1992) seven stage model of coping with TS, Leslie (2005) throws additional light on parents' response to having a child with a medical condition. Having discovered that the child has a medical condition, parents may become even more protective of the young child. The need for protectiveness is obvious to anyone who has parented such children, as they are often ridiculed for their symptoms, or asked to suppress symptoms that may currently be impossible for them to suppress (Leslie, 2005). Having accepted the notion that the child's behaviour is a symptom of a medical condition, the parent may often find that much time and effort must be invested explaining to others why their child cannot do what every other child is doing or why their child should not be punished when other children might be punished for the same activity.

Parents who try to explain to the child's school that these 'behaviours' are really neurological symptoms are generally doing so because of a fear that the child will be blamed for something that the parent has reason to believe that the child cannot control or manage easily. They in instinctive way try to protect their child from a system that tends to punish departures from a fairly rigid set of expectations for how children should behave. Just as some parents may 'medicalise' or 'overmedicalise' behaviours, some teachers attribute too much voluntary intention to the behaviour (ibid). One of the most frequent examples is that of a teacher who, describing a child's tics or compulsions, characterises them as 'attention-seeking' behaviours.

In some cases, then, parents and teachers become polarised in their understanding or explanation of the child's behaviour. Disagreement over the cause of the child's behaviour and whether or not it is a voluntary act is a major source for conflict and disputes between parents of children with TS and school personnel (ibid). Parents are often most concerned about the psychosocial impact of TS, and worry about *'How will other children view my child?', 'Will teachers think he's crazy?', 'Will he be teased because of his tics?'* This is especially so for teenagers who are already struggling with the agonies of adolescence and may feel particular concerns as to how their symptoms may affect their attractiveness to others or their ability to form friendships and romantic relationships.

Many children with TS are teased because of their tics, and some of them are actually rejected because of their tics. Even young children may reject peers because of their tics (Hagin & Kugler 1988). Parents do have cause to be concerned about the child being viewed as less socially acceptable because of his tics (Friedrich et al., 1996). To date, however, there has been little research that has addressed whether an adequate peer education programme could eliminate or ameliorate any negative peer evaluations, although a preliminary study suggests that peer education may be helpful (Woods & Marcks, 2005). This section discussed views and conceptions concerning TS and the process of gradual acceptance of the disability that the parents of children diagnosed with TS usually undergo. The next section discusses the difficulties of children and adolescents who have TS.

2.6 Children and Adolescents with TS

As this research sought to investigate the FQOL of families having a child with TS, this section describes the condition as it appears amongst children and adolescents. Children and adolescents can be categorised as having mild, moderate, or severe TS according to the frequency and complexity of symptoms, and the degree to which they cause impairment or disruption of the person's on-going activities and daily life. For example, extremely frequent tics that occur 20-30 times a minute, such as blinking, nodding, or arm flexion, may be less disruptive than an infrequent tic that occurs several times an hour, such as loud barking, coprolalic utterances, or touching tics (Shapiro et al., 1988). In this sense, there may be tremendous variation over short and long periods of time in symptoms, frequency, and severity. Children with TS may be able to inhibit or not feel a great need to emit their symptoms while at school. When they arrive home, however, the tics may erupt with violence and remain at a distressing level throughout the remainder of the day (ibid). It can be said, then, that the symptoms may be controlled, to some degree, and for a short period of time.

In addition to the moment-to-moment or short-term changes in symptom intensity, some children with TS have oscillations in severity over the course of weeks and months. The waxing and waning of severity may be triggered by changes in the person's life; for example, around the time of holidays, children may develop exacerbations that take weeks to subside (ibid). Other children report that their symptoms show seasonal fluctuation. However, there are no rigorous data on whether life events, stresses, or seasons do in fact influence the onset or offset of a period of exacerbation. Once a person enters a phase of waxing symptoms, a process seems to be triggered that will run its course - usually within 1-3 months (Greene, 1996). Thus, the physiological symptoms found in children with TS may vary in their severity, frequency of occurrence, and intensity.

In addition to physiological symptoms, children with TS show behavioural and psychological symptoms. Children with TS often 'exhibit severe behavioural disorders - intense temper outbursts, non-compliance, volatility, mood instability, and verbal and physical aggression that have the potential to make life extraordinarily challenging and frustrating for them' (Greene, 1996, p. 45). They can also experience a wide range of psychological, behavioural and psychiatric problems in addition to tics. These difficulties also engender interpersonal communication problems (Carter et al., 2000), feelings of anxiety and mood-changes (Robertson, 2002), and may be accompanied by difficulty in concentrating and hyperactivity (ibid) and obsessive behaviours (Robertson & Baron-Cohen, 1995).

These behavioural symptoms may lead parents of children with TS to feel *'uneasily tense and insecure as a consequence of the difficulties of dealing with their child's symptoms'* (Greene, 1996, p. 187), making life very difficult for both the family members and those who interact with them. Research shows that the existence of children with TS in the family influences the relationships between family members (Robertson, 2002). These influences may be expressed in more addiction among family members, more family conflicts, and low quality of interaction between parents and children and higher levels of parental frustration than in families without a child with TS (Cohen et al., 1988; Wilkinson et al., 2001). This is not because daily care for the TS child differs from the care for any other child, but because the symptoms of TS are often combined with related conditions as mentioned before, making daily care harder.

For example, children with TS and ADHD have trouble keeping their attention focused on one activity, so they may need more reminders or supervision when they do daily chores, such as cleaning their room and setting the table. Up to 50% of all children with TS have ADHD .These children have problems with attention span, concentration, distractibility, impulsivity, and motor hyperactivity (Pauls et al., 1993; Robertson, 2002).

Attention problems often precede the onset of TS symptoms and may worsen as the tics develop. Increasing difficulty in focusing attention may reflect an underlying biological dysfunction involving inhibition, and may be exacerbated by the strain of attending to the outer world while simultaneously working hard to remain quiet and still.

Additionally, the symptoms of children with TS who also have OCD may include rituals or routines that interfere with the family's daily routine (Greene, 1996). For example, if a child has a compulsion to wash his hands repeatedly before breakfast, his parents might have a trouble getting him dressed and ready for school in time. The nature of OCD symptoms in children with TS is quite variable. Conventionally, obsessions are defined as thoughts, images, or impulses that intrude on consciousness. Typical OCD behaviours include rituals of counting, checking things repetitively, and excessive washing or cleaning. While many children who have TS do have such behaviours, these symptoms also typify TS itself, therefore these symptoms seems to straddle the border between the tics of the TS and OCD symptoms. For example, children with TS need to touch things a certain number of times, to perform tasks over and over until they 'feel right', and may perform repetitive self-injurious behaviours (Santangelo et al., 1994). OCD, then, is typical of children having TS, but may appear with no regard to TS. A distinction needs to be made here, so that parents and care-takers know how to attend to symptoms with respect to the two disorders.

The need to keep the tics of a child with TS to a minimum by reducing stress can dictate the way the whole family's life is run. It is often found that the family and even the child with TS do not thoroughly understand the range of symptoms involved, nor how they might be expected to handle them (Greene, 1996). For these reasons, TS can cause a great strain on the family as well as on the individual himself.

As noted, parents may find it difficult to accept their child's disorder especially because of guilt feelings associated with the genetic nature of TS (ibid). They must also decipher which behaviours can be controlled by the child and learn how to help the child to do so. In addition, it is difficult to

express the stress involved in dealing with the disorder because pressure and tension may also impact on the child's ability to cope. Thus it is very helpful for parents and the child with TS to learn strategies to cope with this situation (Greene, 1996).

In adolescence and early adulthood, children with TS frequently come to feel that their social isolation, vocational and academic failure, and painful and disfiguring symptoms are more than they can bear (Sukhodolsky et al., 2003). They have difficulty interacting with their peers (Bawden et al, 1998), and lower quality of life (Elstner et al., 2001) than children without TS. A small number of adolescents with TS may consider and attempt suicide (ibid). Conversely, some children and adolescents with TS who exhibit the most bizarre and disruptive symptoms may achieve excellent social, academic, and vocational adjustments (ibid). This means that care and attention should be invested with regard to the specific disorder and needs of children with TS. The next section deals with the integration of the child with TS within the education system.

2.6.1 The Child with TS at School

Because of the centrality of school within children's lives, the child's condition within the education framework is now discussed. Children with TS may exhibit difficulties in daily functioning in school because of the complexity of their symptoms. A study by Piacentini et al., (2003) regarding the level of prevalence of academic problems among children and adolescents with TS showed that 36% (of 200 children aged 6-16) had some sort of academic difficulties at school (Erenberg et al., 1986). Attention problems and hyperactivity can profoundly affect school achievement. At least 50% of children with TS have serious school performance difficulties (Goetz et al., 1999). These difficulties require special intervention, and children with both TS and Attention Deficit/ Hyperactivity Disorder (ADHD), are especially vulnerable to serious, long term educational impairment.

In their research sample, Goetz et al. (1999) found that 52% of the children with TS seen in their clinic experienced social or educational dysfunction. Of these, 39% required special education placement, 10% had been retained at least 1 year in school, and 29% experienced disciplinary problems. Whereas half the sample experienced significant social and educational dysfunction as children, only 32% of the same sample experienced social or educational dysfunction as adults. On a positive note, all the sample participants had completed high school and at least two years of college (adult sampling was conducted while the individuals were in their 20's). The researchers found that 71% of the adults studied were currently employed or pursuing their education. Of the adults who had social or educational dysfunction as children, 50% had social or educational dysfunction as adults; although 13% of children who had not previously experienced education or social dysfunction went on to develop social or education dysfunction as adults. While the majority were at follow-up doing well and were well-integrated in their communities, over one quarter of the adults were disabled with problems that included alcohol abuse, unemployment or criminal activity. The fact that these measures did not correlate with tic severity measures but did correlate with early childhood dysfunction suggests that early and effective interventions for co-morbid conditions (perhaps even more so than for tics) may impact on the prevention of dysfunction in adulthood. It can be said, then, that while children who have TS may experience social difficulties at school, and might dysfunction as pupils, their academic future is not necessarily grave.

The negative impact on the child's functioning at school is obvious, particularly when the child with TS also has ADHD. This is because the child with TS and ADHD is characterised as having difficulties with self-regulation, including attention difficulties, hyperactivity and impulsivity. They also have many social and educational difficulties and find it hard to adapt their behaviour to the environment (Greene, 1996).

Lots of things can happen at school - being teased by other children, feeling socially isolated or rejected, feeling frustrated and embarrassed over struggles on certain academic tasks, being misunderstood by the teacher - to fuel meltdowns at home (ibid, p. 266-267).

It appears that social problems caused by TS may entail educational problems and dysfunction at school.

Some children and adolescents with TS (percentages vary greatly in different studies) have significant problems with labile emotions, impulsivity, and aggression directed to others. Temper fits that include screaming, punching holes in walls, threatening others, hitting, biting, and kicking are common in these people (Carter et al., 2000), especially when they also have ADHD, which makes impulse control a considerable problem. At times, the temper tantrums can be seen as reactions to the internal and external pressures of TS. A specific aetiology for such behavioural problems is, however, not well understood.

Nevertheless, these phenomena create much consternation in teachers and great anguish both to those with TS themselves and to their families (ibid). The treating physician or counsellor is often asked whether these behaviours are involuntary, as tics are, or whether they can be controlled by the person with TS. Rather than trying to make such a distinction, it is perhaps more helpful to understand whether such people experience themselves as being out of control.

Management of such behaviours is often difficult and may involve adjustment of medications, individual therapy, family therapy, or behavioural retraining. The intensity of these behaviours often increases as the tics wax and decreases as the tics wane (Pauls et al., 1993). It seems that aggression directed at others, whether controlled or involuntary, together with ADHD disorder, tends to determine the reaction of the environment towards children with TS.

2.7 Siblings of Children with TS

Research shows that the experience of growing up with a sibling with a disability can be strenuous and influential (Bawden et al, 1998), presenting specific challenges and emotions which must be coped with. One of the most common challenges in the case of a sibling of a child with TS is dealing with feelings of 'embarrassment' and 'shame'. (ibid; Shimberg, 1995)

Sometimes, the siblings of children with disability feel that they are trapped on both sides: On the one hand, they live in the world of their 'normal' peers, yet on the other hand, they have one foot within a family with a disability. Many parents of children with disability express sorrow that their healthy children are neglected because of the disability within the family. Parents feel that their children have been deprived of an opportunity for happiness and the pleasure of having a healthy sibling, and their right to have a worry-free childhood (Pezerstone, 1987; Levy-Shiff, 1999). Furthermore, parents are pressured by their worries, concerning both their healthy children and children with disabilities and may encourage their healthy children to express positive feelings and behave lovingly towards their sibling with a disability. To a lesser degree, they encourage expressions of anger, jealousy, isolation or embarrassment on the part of their healthy children (Pezerstone, 1987; Levy-Shiff et al., 1998; Levy-Shiff, 1999).

The attitudes of some siblings towards themselves, their parents and their sibling with disability are affected by this approach of parents. Even in sensitive and supportive families, it is impossible to avoid feelings of deprivation and bitterness, guilt and shame on the part of healthy children towards their sibling with disability (Eylon, 1983; Shulman et al., 1995; Shulman et al., 2001). Therefore, apart from the parents themselves, other family members, and specifically brothers and sisters of the children with disability, are faced with a number of emotions: fear, anger, jealousy, isolation, guilt feelings, identification, confusion and shame.

Siblings of children with disability, like their parents, deal with the situation with a wide variety of emotions, both positive and negative (Eylon, 1983; Levy-Shiff et al., 1998). The siblings of children with disability must often live alone with these feelings, and do not usually share their feelings with their parents or their environment, (Weiskopff, 1990; Feigin, 2002).

As a result, two risks arise: First of all, there is concern that the healthy child will become a target for high expectations of parents who want to compensate themselves for the disappointments and frustrations involved in raising the child with a disability (Weiskopff, 1990; Feigin, 2002). For example, they may pressure the child without a disability to succeed socially and academically. The healthy child, who tends to keep his emotions to himself, may not say anything about this and may spend his life trying to fulfil his parents' expectations. Secondly, there is a risk that the development of the child without a disability will be harmed as a result of neglect and lack of opportunity to create social ties outside of the family, because of the great deal of time and care that the parents invest in raising the child with a disability. Therefore, it is clear that the siblings of children with disability go through a process of adjustment to the disability, and require time to accept this fact, resembling the process that the parents themselves undergo (Weiskopff, 1990). According to studies conducted in Israel, it seems that raising a physically or mentally disabled child is a significant source of pressure for the entire family (ibid; Brown *et al.*, 2003).

TS involves external physical forms of behaviour, which are not controlled and are very exceptional, and consequently draw unsympathetic attention from the environment. Thus, siblings of children with TS, who like other children want others to believe that their family is 'normal' in every way, and not different from other families, tend to feel great shame when they are in social situations with their siblings. Furthermore, they tend to detach themselves from their friends or avoid inviting friends to their houses. These feelings of embarrassment and shame become even stronger during adolescence, when siblings of the child with TS feel an acute need to fit in and confirm with their environment (Buell et al., 1999).

These responses of embarrassment and shame are very natural for siblings of children with TS. Nevertheless, sometimes these feelings may cause the sibling to join with his friends in ridiculing their sibling's behaviour. Consequently, it is important for the parents of the child with TS to accept the feelings of their other children and provide them with room to express and discuss their feelings with them (Shimberg, 1995). It should be noted that despite feelings of shame and embarrassment, sometimes siblings of a child with TS feel profound 'relief' when they find that the child's strange behaviour has a scientific name and particular reasons, and that their sibling actually cannot control this behaviour. This relief also stems from their ability to stand up to the teasing of their friends, by explaining that the sibling's behaviour stems from a neurological problem which cannot be self-controlled, (ibid).

Another emotional difficulty which siblings of a child with TS may have to face is 'a feeling of guilt'. Shimberg (1995) found that siblings of a child with TS, like siblings of children with other disabilities, tend to feel guilty, thinking that they had caused the disorder as a result of their expression of anger towards the sibling or even because they had hoped that something bad would happen to the sibling. Interviews with the brothers of children with TS revealed that one of the main reasons for guilt feelings among siblings is that they tend to feel jealousy towards their brother/ sister for the attention that they receive from their parents over the years, before diagnosis. And after finding out, after diagnosis, that something is wrong with their sibling, they tend to feel great guilt, anger and even jealousy (ibid). The feeling of guilt and shame evoked by siblings towards their brothers or sisters who have TS is in line with the same feelings experienced by parents of such children, thus influencing the family as a unit. Haerle (1992), confirmed that siblings of a child with TS often deal with feelings of 'anger' and 'jealousy' towards the sibling for taking most of their parents' attention. According to Shimberg (1995) the jealousy of a sibling towards the child with TS arises whether the attention that parents pay to the affected child is positive or negative. She argues that many siblings report that they themselves 'get into trouble', and show strange behaviour, even performing similar tics, only to win attention from their parents, even if it causes them to be punished.

Furthermore, interviews (Shimberg, 1995) show that the siblings of a child with TS often felt anger towards themselves because of their lack of loyalty towards their brothers, a feeling that they may live with for many years.

Another challenge facing siblings of a child with TS is their 'identification' with the sibling who has the disability. As previously mentioned, sometimes one may see the sibling of a child with TS presenting similar behaviour and tics, in an attempt to win their parents' attention. According to Pezerstone (1987), identification with the sibling with disability can lead the healthy sibling to mimic the strange behaviour, and to develop similar symptoms.

A further challenge for siblings is the need to deal with 'fears'. Haerle (1992) argues that siblings of children with TS sometimes fear for their own health. This fear becomes stronger when the aetiology of the syndrome is understood. As mentioned above, one of the causes of TS is a genetic fault. According to Shimberg (1995), children whose siblings are disabled are afraid to 'catch' the condition. In the case of siblings of a child with TS, the syndrome may not be 'contagious', but it is possible for a sibling to develop the condition in the future, particularly if they are still young. The fact that TS is genetically based creates a realistic fear among siblings, and also the fear of becoming parents to TS children themselves. Many siblings report that they are afraid that they carry the gene and may transmit it to their own children (ibid).

Two more challenges faced by siblings of disabled children, in particular among families of children with TS are: 'fighting and aggression', and 'distracting symptoms'. One of the specific problems of healthy brothers growing up alongside a sibling with TS is the great degree of frustration regarding their sibling's strange behaviour, often causing them to fight with and even to beat up their siblings to make them to stop the strange tics (Shimberg, 1995). This is especially evident among siblings with TS with coprolalia tics (involuntary rude remarks, sometimes yelled), caporpraxia tics (rude gestures), or particularly aggressive tics. The daily tension created by growing up alongside a sibling who exhibits tics often causes healthy

children to get into fights with their siblings with TS and to blame them for various things, even if these are unconnected with the syndrome (ibid).

Another problem is that the tics and behaviours of the siblings with TS are very distracting, and it is difficult to ignore them. For example, a child with TS may have vocal tics that include making loud noises, or may also have OCD, leading him to open and close doors in the house, over and over again (Shimberg, 1995). These phenomena are difficult for the entire family and healthy siblings within the family, who are sometimes trying to do their homework or watch television, or be alone in their rooms, so that they find it especially straining to live in a house with a child with TS.

In conclusion: like their parents, siblings of children with TS must deal with a variety of challenges and difficulties and must go through a process of adjusting to their sibling's disorder. Despite these difficulties, Robertson and Baron-Cohen (1995) found that siblings of children with TS tend to develop a deeper understanding toward their brother or sister, and an understanding of medical conditions in general. They also tend to show more understanding and esteem for good health and a healthy lifestyle. This is in keeping with earlier theory and research on the subject of siblings of a child with disability, which found that the siblings developed from a status of stress, crisis and pathology, to greater understanding towards brothers and sisters with disability (Glidden et al., 1992).

2.8 Summary regarding the Family of the Child with TS

Like other families who deal with a child with disability, families of children with TS also undergo crises and changes in various areas of family life. Those changes may appear in the family as an organisation and a system, changes in systems of relations between parents and children and also within the parental relationship. There have been few studies specific to families of children with TS in Israel; however, it was found that the birth of a child with TS in some cases creates a crisis situation for the family which has many implications for the family's lifestyle (Levy-Shiff, 1997; Levy-Shiff et al., 1998). Many studies indicate that the family is an important mediating factor in the emotional and social adjustment of the child. For this reason, information regarding the manner in which parents and siblings of children with TS tend to respond to this crisis situation and their coping strategies, can help professionals in the mental health field, doctors, and neurologists who encounter such families in the future. It can also help the families themselves to better understand how the birth of a child with TS affects the family, and help them learn better ways of dealing with the many difficulties that arise while raising a child with TS together with their other children in the family. The next section discusses conceptions of FQOL in relation to families who have a child with TS.

2.9 Conceptions of Family Quality of Life

2.9.1 Preview

This section attempts to provide a comprehensive explanation of the concepts of Quality of Life (QOL) and FQOL. The variables of FQOL, which are the main research variables are defined and explained. Additionally, this section discusses various definitions and indicators of the family in Israel. Despite a long history of QOL studies, most research efforts have focused on conceptualisation of and measuring individual QOL. It is only recently that FQOL has started to draw attention. Therefore, the conceptualisation of FQOL depends much on the literature about individual QOL.

2.9.2 The Modern Family

Rapid social changes in recent decades have meant that traditional sociological definitions of the family which were accepted in the past have lost their validity. It has become difficult to achieve a consensual definition (Lavee et al., 1996). Bar-Yosef (1996) argues that sociological research of the family is in the midst of a theoretical crisis, the main signs of which are: doubts regarding the importance of the family, considerations regarding the boundaries of families, and a transition from precise and clear definitions of family to definitions which are open and vaguer.

Despite these difficulties, several attempts have been made to create modern definitions of the family. Peres and Schrist (1978) argue that the most widely accepted definition of family in industrialised countries is '*a post-nuclear family system*', whose main characteristics are: 1) individuals belonging to a family are more autonomous and less mutually dependent; 2) values of conformity and obligations towards others are replaced by values of self-fulfilment; 3) the stability of the family is declining; 4) a transition of roles from the family to other social systems; 5) values of equality have entered the family and have made dominance and authority relationships unacceptable, both between the sexes and between generations; 6) alongside the conventional family, other alternative family types are

developing, such as: two-career families, single-parent families, and single-sex families.

Bar-Yosef (1996) argues that modern definitions attempt to deconstruct the classical model of the family into three components, which may overlap to various degrees, i.e. intimate relations, parenting, and the household. Modern definitions of intimate relationships attempt to provide room for all possible relationship types and describe a reality in which there is a continuum of various types of intimate relations, beginning with formal, established models and up to and including intimate relationships with no commitment and lacking stable boundaries of time and space.

According to Bar-Yosef (1996), this engenders questions regarding the conditions which are sufficient and necessary to define intimate relationships. Difficulties in defining parenting relate to the distinction between two types of parenthood - genetic and functional, and also the dissolution of the connection between intimate relationships and parenting, which, in the past, was perceived as necessary. Modern fertilisation technology has raised a new group of problems in defining parenthood. Addressing the household component when defining the family is also at question today, as intimate relations and parenting do not necessarily require living together. This raises questions regarding the essence of continuing relationships between adult individuals, and the manner in which it is possible to define them.

Most researchers share the opinion that changes and developments in the structure of the family and the study of the family (Peres & Katz, 1991; Sharlin & Shamai, 1995) are closely and mutually related to changes in the society in which the family exists (Bar-Yosef, 1996). Albeck (1990) summarises the main social processes effecting changes in the family, and which she also argues undermine its strengths, under two subheadings: 1. industrialisation and urbanisation processes; 2. changes in social values and the rise of values of secularism, democracy and liberalism-individualism. She argues that industrialisation and urbanisation processes have led to a decrease in family functions and the expansion of families' relations and

dependence on entities external to the family. In addition, a change in women's status, delayed age for marriage, and declining birth rates have limited the social roles of the family and transformed the family into a unit that depends mainly on emotional ties and expectations of happiness. Trends towards secularisation and democracy have led to a weakening of supervision by religious institutions and moral norms, and stronger emphasis on the individual's happiness and freedom, at the expense of adjustment and conformity to the demands of a traditional family lifestyle. Ideals of innovation and change, and orientation towards the future within modern society also contribute their part to weakening the status of the family, which is related, among other things, to awareness of the past, tradition, and continuity.

It appears that changes in society affect the family the same way as changes in the perception of family affect society. Just as the family is affected by social changes, changes occurring within the family affect society at large (Diomon, 1995). In light of this mutual interdependence, any analysis and definition of the family must consider the family's social and cultural context (Sharlin et al., 1991; Diomon, 1995). This thesis deals with the specific situation of a group of Jewish families in Israel who have a child with TS. Although definitions of the 'family' from other countries may be useful in clarifying the situation of the Israeli family, nevertheless the context of the Israeli Jewish population has its own specific characteristics. Therefore, before discussing the results of various studies regarding the Jewish family in Israel, an attempt is made to define the Israeli Jewish family and its unique attributes.

2.9.3 The Family in Israel

Demographic data indicate that Israel is more 'family oriented' than Western and Eastern European countries, according to the following indices: a relatively high birth rate, a relatively low divorce rate, and few births outside marital relationships (Katz & Peres, 1986; Katz, 1998, 2000). The Israeli Jewish family seems to have remained relatively stable despite the impact of industrialisation and modernisation in Israel. Katz & Peres (1986) offered a

number of explanations for this fact: 1) the tense security situation in Israel and the fact that Israel is a 'country under siege', have led to a higher Israeli awareness of risk, and this strengthens family relations; 2) a large and growing part of the Israeli Jewish population are of Oriental descent (originating from Africa and the Middle East), typified by more traditional family models; 3) the central role of religion and tradition in Israel, as expressed on an institutional and moral level; 4) the fact that Israeli society is fairly small and intimate, which serves as an effective supervisory means, increasing the individual's relationship to his family.

The validity of these explanations by Katz and Peretz (1986) regarding the relative stability of the Israeli Jewish family was reinforced by the work of Sharlin et al., (1992), and constituted the basis for surveys performed by later authors (Albeck, 1990; Katz & Peres, 1993). Now, it may be necessary to examine additional or alternative explanations for this phenomenon. For example, Albeck (1990) notes the explanation of Marcus and Doron (1988), who argue that the strong dependence of young families in Israel on the financial support of their families of origin, contributes to strengthened intergenerational family relationships. Safir (1993) argues that the stability of the Israel Jewish family is supported by the patriarchal values, of Jewish tradition. He also draws attention to the high degree of importance placed by Israeli society on the issue of fertility, in particular as a result of the 'demographic problem', and the desire to maintain a Jewish majority in Israel. This argument also holds true of course for the aspirations for a demographic majority of the Arab population of the State.

Safir (1993) also lists several examples of the centrality of the family in the Israeli Jewish experience, including the tendency of young families to own their own flats and to maintain a stable place of residence, generally in the vicinity of the extended family; the flourishing wedding ceremony industry and the social requirement, mainly (but not only) directed to the women, to continue to care for their children even after the children leave home, and later to take care of their grandchildren and their aging parents.

Despite the fact that the Israeli Jewish family has been relatively stable, over the past few decades, there have been some, not very dramatic, but consistent changes. These are expressed in a rising divorce rate, an increasing rate of births out of wedlock, an increase in age at marriage, and a decrease in the birth rate. Furthermore, new family models and lifestyles are developing alongside the traditional family model. There has been a significant rise in the percentage of single-parent families, in the percentage of second marriages creating 'step' or 'blended' families, and the percentage of couples living together without marriage as a precursor to marriage (Katz & Peres, 1993). These trends point to an on-going change occurring in the perception of the family unit in contemporary Israeli society.

An additional demographic influence arrived with the waves of Jewish immigration from Russia and Ethiopia, relating to the structure of the family and its problems, and these waves have significantly raised the number of single-parent families in Israel (Sharlin et al., 1992; Katz & Peres, 1993). With increased globalisation, it is reasonable to hypothesise that the changes in the structure of Israeli Jewish families will continue, and they will become more similar to families in other modernised industrialised countries. Nevertheless, there are other trends which resist these changes in the traditional family, inclining towards religious conservatism, indicating that the family may still maintain its relative stability in the near future (ibid; Sharlin & Shamai, 1995).

Just as the Israeli Jewish family has undergone changes over the years, sociological research concerning the family has also altered its focus. Katz & Peres (1993) surveyed research on Jewish families in Israel, beginning from the 1950s to the 1980s. They explained that research of the 1950s and 1960s tended to see the family (e.g. kibbutz families) as reflecting and dealing with the personal and social needs of individuals and groups, and the absorption difficulties of the immigrant 'melting pot'. Later studies related to internal family components, such as the struggle for sexual equality in the family, the increase in single-parent families, and the impact of mothers' employment outside the home on the family.

According to Katz & Peres (1993), this trend shows that with the years, Israeli research has begun to focus on the same issues as family research throughout the Western world.

Sharlin et al, 1992 surveyed studies regarding Israeli Jewish families in which the dependent variables were various indices of FQOL or individual QOL. Welfare, health, satisfaction and adjustment were only some of the concepts mentioned in this context. In their discussion on the definition of and measurement of quality of marriage, they noted that despite the many years of research dedicated to this issue, there is still much conceptual confusion and disagreement regarding methods of measurement (Sharlin et al., 1992). Apparently, this statement is no less valid, and perhaps even more valid, when attempting to discuss the QOL of an entire family.

For the purpose of this research, the family is defined as a social institution comprising a mother and father plus children who are living at home or a single parent with children living at home. The research related specifically to the family that includes one or more children who are diagnosed as having TS and live at home

2.10 FQOL

Relevant research and theoretical literature indicates that FQOL is difficult to define (Turnbull, et al ,2000) Despite the difficulty involved in defining FQOL, there are a number of aspects of FQOL that are agreed upon worldwide (Browne et al., 1997), aspects that appear to varying degrees and are related to various family experiences. For example, families hope to have children and raise them according to the familial and social conventions of a given society. They want to be healthy and to function in a perfect manner within the family and outside of it (ibid). Families like to attain personal and group achievements and like to do things together as a group, to play, to laugh, to create and to have fun (ibid; Isaacs et al., 2007; Burgess & Gutstein, 2007).

According to Browne et al. (1997) families enjoy experiences within the society in which they live and prefer family members to remain close and connected, loving and considerate towards one another. They like to be in contact with other families and to continue existing customs passing on experiences, dreaming and aspiring to connect with powers that are stronger than them. They prefer financial, psychological and social stability. In the event that families do not achieve their desires, this may cause a lack of satisfaction, a lack of self-confidence, increased tension and opposition (ibid).

Although each family understands the concept of quality of life differently, there is a general understanding regarding the concept. Families express satisfaction with their quality of life when they achieve what all other families worldwide are interested in achieving and when they are able to empower themselves and their lifestyle (ibid). This definition seems vague, because it lacks any specific information about criteria or domains of satisfaction.

Despite this fact, no two families are exactly the same and every family is its own unique unit, functioning individually, liking and hating different things, and very unique. This makes it difficult to define FQOL. When defining FQOL, aspects that are important to all families should be considered,

although it is also necessary to relate to the uniqueness of each family and of the individuals within the family. Therefore, when attempting to define FQOL the different outlooks that families have regarding themselves, the variables involved in concept of 'a good life', and also the values of the particular family should be understood.

2.10.1 The Definition of FQOL

As noted above, researchers have for many years studied the QOL of individuals within the family, and have only recently begun to pay attention to the issue of FQOL (Turnbull et al., 2000; Isaacs et al., 2007; Burgess & Gutstein, 2007; Jackson et al., 2009). Therefore, the definition of the concept of FQOL relies on many definitions developed with regard to individual QOL.

Any discussion of QOL usually refers to how 'good' life is. If human beings are seen as organisms with 'potential', capable of mental and social self-realisation, then the purpose of life is the ability to allow this 'potential' to develop in both an individual and ecological context. Therefore, QOL, or in other words, 'a good life', means the ability to maximise life's 'potential' in a particular social and ecological context.

QOL is shaped by a variety of life conditions that interact in complex ways. What is considered to be a 'good' life is different for different people. People in various parts of the world have defined quality in their lives in rather distinct ways. Different groups and individuals often think of QOL in ways that are specific to their own life situations and their own characteristics (Browne et al., 1997). But a slightly more profound examination of this issue reveals that it is not always a simple task to determine how 'good' life is or what the meaning or purpose of life is (Turnbull et al., 2000). QOL relates to the person's general expectations of life, therefore, its conceptualisation varies according to individual perceptions.

In many cases, QOL relates to a cognitive judgment regarding satisfaction with life and an affective assessment of emotions and moods. It relates to individuals' overall assessment of their internal experiences, including

thoughts, attitudes, motives, dreams, and emotions (Diener & Lucas, 1997), and their subjective and intuitive assessment of the quality of their experience as they are able to rationally and responsibly report. QOL may not be explainable solely on the basis of objective situations that the individual experiences as it is affected by the individual's personal perceptions and attitudes (Pavot & Diener, 1993), and therefore, different people will not necessarily assess the same life circumstances in the same way.

In an historical perspective, the definitions of QOL have changed over the last decades, and the term has been defined differently by different researchers. Thus, QOL has been defined as the difference between the hopes and expectations of the individual and their present experience (Calman, 1984), while others see QOL as a sense of personal satisfaction that is more than contentment and happiness but less than fulfilment (Coulter, 1990). It is experienced when a person's basic needs are met and when he or she has the opportunity to pursue and achieve goals in definitive life settings, namely, work, school, home, and community, (Goode, 1988).

Although QOL is defined in different ways, researchers have agreed on two fundamental concepts: QOL is multi-dimensional, and it includes both subjective and objective dimensions (Halpern, 1993; Testa & Simonson, 1996). Thus, in QOL research a distinction is often made between subjective and objective QOL (Quality-of-Life Research Unit, Toronto, 2001). Subjective QOL is about feeling good and being satisfied with things in general. Objective QOL is about fulfilling the societal and cultural demands for material wealth, social status and physical well-being.

QOL is also the degree to which people enjoy the important possibilities of their life (Oullette-Kunts & McCreary, 1996). Possibilities result from the opportunities and limitations that people are offered in their lives and reflect the interaction of personal and environmental factors. QOL includes, but is not limited to, the social, physical and emotional functioning of the child and adolescent, and when indicated, their family members, and it necessarily alters according to changes that occur throughout the child's development.

QOL is measured by the difference, at a particular period of time, between the hopes and expectations of the individual and the individual's present experience. It is influenced by the individual's past experience, present lifestyle and personal hopes and ambitions for the future (Calman, 1984).

As mentioned earlier, this research sought to explore the perceived FQOL of families who have a child with TS, with the aim that this exploration and the insights gained from this process might inform best practice and policy makers who treat and determine policy for these families. FQOL is defined as *'a dynamic sense of wellbeing of the family, collectively and subjectively defined and informed by its members, in which individual and family-level needs interact'* (Zuna, 2009).

Using cautious inference from the descriptions and principles used in the literature for the measurement of individual QOL, FQOL measures the degree to which families have meaningful life experiences that they value. FQOL measurement can serve as an impetus to guide families to move toward a meaningful life that they can enjoy and value, and measures the degree to which life's domains contribute to a full and interconnected life. FQOL measurement is undertaken within the context of environments that are important to families: where they live, work and play. It is important to mention that FQOL measurement is based upon both common human experiences and unique, individual family experiences.

Research on the subject of FQOL is still in its infancy. To date only a few studies have been conducted on the subject (Baxter et al., 2000; Brown et al., 2003; Turnbull & Turnbull, 2001; Turnbull et al., 2003; Turnbull et al., 2004; Zuna, 2009).

There has been increased interest in research on the topic of families, and how they are affected by the presence of a child with a disability within the family (Hawkins et al., 1993; Lavee et al., 1996; Duvdevani et al., 1997; Baxter et al., 2000), but the issue of FQOL has not been studied until quite recently. Researchers found that the presence of a child with a disability in the family affects the interpersonal relations within the family (Glidden, 1989; Cummins & Baxter, 1997). It is well known that when relations are fulfilling,

everything functions well, and this affects both the QOL of the individual within the family, and the family as a unit. Similarly when one family member has a certain disability, and relations are undermined or tension is caused in the family, this may negatively affect both the individual within the family and the family unit (Glidden, 1989; Cummins & Baxter, 1997). It follows, then, that the existence of a child with TS in the home might affect the family members' perception of the FQOL. This section has discussed how FQOL is perceived by the research literature. The next section delineates in more detail the domains that comprise FQOL.

2.10.2 Domains of FQOL

This section discusses the domains of FQOL described in the relevant research and theoretical literature, and their relevance to the current study. A group of researchers from the United States (Poston et al., 2003) created the foundation for the concept of FQOL, employing a qualitative research programme in order to develop grounded theory that would conceptualise FQOL domains and indicators. They defined FQOL as conditions that meet the family's needs, in which family members enjoy their life together as a family, and family members have the chance to do things that are important to them (ibid). They focused on families which included one member who had a disability, (Brown et al., 2003).

These investigators of FQOL, identified five main domains of FQOL based on extensive qualitative (Poston et al., 2003) and quantitative (Brown et al., 2003; Hoffman et al., 2006) analyses. Their findings formed the basis for a pilot version of the Beach Centre FQOL Scale. Park et al. (2003) conducted an initial development and psychometric evaluation of the pilot FQOL scale. After further evaluation of the factor structure of the scale, they identified five uni-dimensional FQOL domains: Family Interaction, Parenting, Emotional Well-being, Physical and Material Well-Being and Disability-Related Supports. The FQOL scale measures family members' perceptions of the importance of those domains and their level of satisfaction regarding the domains.

Family Domains

This thesis focuses on the family domains of FQOL. Therefore, the literature on the following five family domains is reviewed below: family interaction, parenting, financial well-being, emotional well-being and support for family member with a disability.

Family interaction

Family interaction means that the *'family enjoys spending time together and is able to handle life's ups and downs. Family members support each other to accomplish goals and talk openly with each other. They solve problems together and show that they love and care for each other'* (Turnbull et al., 2004, p.388-392).

Family interaction means the relationships among family members and the relational environment in which the family operates. Family interaction includes family routines, relationships among family members, roles, emotional climate, communication and behavioural discipline (Park & Turnbull, 2001). Communication was found to be one of the most salient ways to counteract stress in families, especially when there is a child with a disability (Turnbull & Turnbull, 1990). One of the most powerful predictors of positive sibling adjustment when growing up with a brother or sister with a disability is communication (Gold, 1993). Parents who were knowledgeable about their child's disability were better able to provide accurate and essential information regarding the sibling's disability (Turnbull & Turnbull, 1990), and were also more successful in fostering positive coping skills in their children with disabilities (Gamble & Woulbroun, 1993).

The fact that a child in the family has a disability can impact on the family in negative and positive ways. Negative impacts may include an extensive amount of time devoted in taking care of the child's special needs, which reduces the time for family members to spend time together, limited communication among family members because of the child's lack of communication skills and additional responsibilities for siblings in the family (Krauss, 1993; Blacher, 1994).

Having a sibling with a disability can present unique challenges. Attention and energy can sometimes become too focused on the child with 'special needs'. Given that siblings of children with disabilities mirror the attitudes of their parents (Gold, 1993) the most powerful predictor affecting sibling acceptance of a child with a disability was found to be parental attitude, in particular, the attitude of the mother (Olsen et al., 1999). An optimistic and accepting parental outlook was positively correlated with improved personal adjustment in siblings without disabilities (Powell & Gallagher, 1993), as well as enhanced relationships with their siblings with disabilities (McHale & Gamble, 1989).

However, having a sibling with a disability may have positive, as well as negative, effects. researchers studying siblings of children with a wide range of disabilities have found no differences in self-concept or in perceived competence between groups of children who do and do not have a sibling with a disability (Roeyers & Mycke, 1995; Fisman et al., 2000; McMahon et al., 2001; Singhi et al., 2002). College-age siblings of individuals with disabilities had a higher internal locus of control than did their peers (Burton & Parks, 1994).

Having a brother or sister with a disability does not necessarily cause maladaptation or pathology in children, and few children seem to be harmed by the experience (Rossiter & Sharpe, 2001). Most relationships between children with disabilities and their siblings are positive, nurturing, and satisfying (Kaminsky & Dewey, 2001). There is even evidence that sibling relationships involving a child with a disability may be more positive than those between comparison siblings who have no disability (Cuskelly & Gunn, 2003). Non-disabled siblings have more difficulty creating play accommodations that lead to sustained interaction when children's disabilities are more severe or include social deficits, and when children with disabilities engage in behaviours that disrupt play (Heller et al., 1999). Siblings of children with disabilities frequently assume dominant roles as they help, teach, and manage their brothers and sisters. Both older and younger siblings of children with disabilities assume expanded childcare roles as compared to their peers (Cuskelly & Gunn, 2003). The social

interactions of younger siblings of children with disabilities are characterised by younger sibling role dominance; comparable interactions between non-disabled siblings follow a more normative pattern of older sibling dominance (Stoneman & Berman, 1993).

Siblings of children with disabilities who received more time and attention from their parents reported warmer sibling relations but perhaps surprisingly increased adjustment problems. In contrast, siblings of non-disabled children who received more time and attention reported poor sibling relations but less adjustment problems (McHale & Pawletko, 1992). McHale and Pawletko (1992) suggested that more favourable parental treatment of siblings of children with disabilities induced positive self-regard, yet it also engendered negative feelings toward their brothers or sisters with disabilities thus creating the adjustment problems, which in turn, created feelings of anxiety and guilt, eventually resulting in increased compassion and kindness within the sibling relationship (*ibid*).

Open, honest communication was found to provide assurance and comfort to siblings (Turnbull & Turnbull, 1990) and to assist siblings with their feelings of guilt, anger, resentment, and jealousy (Gibbs, 1993), as mentioned before. Straightforward communication helps families to gain a practical and shared set of assumptions and meanings about themselves in relation to one another; and about their family in relation to the community and society. These shared meanings reduce individual uncertainty about the disability and make coordination of a collective family response possible. A coordinated response results in greater stability and cohesiveness among family members (Patterson & Garwick, 1994).

As noted above, recent research reports some positive contributions that the child with disabilities makes to family interaction. The child with disabilities may become a source of family unity and closeness, if the family members increase their tolerance and understanding toward each other and siblings show a higher level of internal locus of control (Burton & Parks, 1994; Turnbull & Turnbull, 2001). Studies show that positive changes in parents' interaction behaviours impact on child-parent relationships (Hemmeter &

Kaiser, 1994). Early intervention affected child development when mothers changed their style of interaction toward higher responsiveness to the child's behaviour (Mahoney & Kim, 2004). Siblings of children with disabilities reported that they helped their disabled sibling more than they played with them and that they expressed more positive affect toward their siblings with disabilities than they did toward non-disabled siblings (Miller, 1978). In sum, consideration of family interactions as a domain in FQOL indicates that any change in these interactions affects all the members of the family, hence its significance and relevance to this study.

Parenting

In this thesis, parenting means that *'adult family members help children with schoolwork and activities. Adults know other people in the children's lives, namely, friends and teachers, and teach the children how to get along with others and how to make good decisions. Adult family members have time to take care of the individual needs of every child and help children learn to be independent'* (Turnbull et al., 2004, p.388-392). Parenting focuses on the responsibility of a parent toward a child in the family, including teaching, guiding and discipline.

Research about parenting children with disabilities has often focused on the negative aspects, such as family disruption and stress. However, many researchers suggest that the negatives have been overstated and the positives overlooked (Sandler & Mistretta, 1998). While families with children with disabilities do experience challenges, newer studies show that with good support they can be as vibrant and healthy as families without children with disabilities (Peck, 2002).

Research show that some parents feel their families have been blessed because of their special child. They say they are closer and have become more compassionate, tolerant, sympathetic, flexible, and selfless. While each family experiences struggles unique to their child's disability and family situation, the families who report a positive parenting experience tend to share several characteristics in common (Li-Tsang et al., 2001).

Discussions and sensitivity are needed to examine the different ways parents might choose to react. Discussions can actually help to sustain a strong relationship between parents. Life's daily challenges become more bearable and this marital bond also helps children cope. Families that deal well with a child who has special needs keep a positive attitude, have realistic expectations of their child, and are less preoccupied with negative thoughts (Yau & Li-Tsang, 1999).

Religious faith and beliefs provide many parents with strength, support, and hope during times of difficulty (Olson et al., 2002). Belief in a divine plan allows parents to find meaning in their situation and to rise above discouragement. Many parents see their child with special needs as a child of God and believe that they have been blessed to be entrusted with this special spirit in their home. They feel their child was sent to them for a purpose (ibid).

There are positive relationships between parenting and marital quality and social support, and negative relationships between parenting and stressful events (Bradlyn et al., 1996). The severity of the child's disability has also been found to be correlated to parenting, in that the more severe the child's disability, the less responsive and adaptive parenting styles will be (ibid).

Parents can move to more adaptive parenting styles when organisation and support is provided from multiple systems (Osofsky & Thompson, 2000). Support groups and parent education programmes have a positive impact on parenting (Turnbull et al., 2000). Parents who participate in a programme for improving parents' self-efficacy, effective child raising, and problem-solving skills, show significant increases in positive parenting (Miller et al., 2001). Parental influence on sibling outcomes is an interactive process by which individual children are socialised according to parental child rearing strategies, which, in turn, predict the relationship between siblings (Stoneman & Berman, 1993).

Some mothers of children with ADHD have been found to display negative and disapproving reactions to their child's disruptive behaviour and these are typically reprimanding and controlling mothers (Cunningham & Barkley,

1979; Mash & Johnston, 1982; Befera & Barkley, 1985). Yet, it is difficult to determine whether mothers' disciplinary actions are merely a reaction to or a cause of the child's deviant behaviour (Fisher et al., 1986). Mothers of children with (ADHD) often face public accusations of blame, as well as personal feelings of responsibility for their child's disorder (Bennett, 2007). Within social contexts, the associated behaviours of children with (ADHD) were found to cause mothers a great deal of embarrassment and stigma, feelings of isolation, failure, blames and negative self-esteem, (ibid). It was also found that mothers of children with disabilities experienced depressive symptoms almost twice as often as mothers of children without disabilities. Although these mothers were in the minority, their depressive symptoms often negatively influenced their parenting style and limited their ability to socialise the sibling relationship (Stoneman & Berman, 1993). When mothers were more negative, conflict between children with disabilities and their siblings was higher, as was sibling depression and anxiety.

Further evidence that parents structure the contexts in which young siblings interact and provide feedback to their children as to which behaviours are desirable or undesirable (ibid) has been found by research in this area: mothers' structuring of play and encouragement of sibling interaction may be disrupted when the family climate is negative and conflict-filled (Dallas et al., 1993). Depressed mothers were inconsistent in disciplining their children and used verbal and physical punishment more frequently than non-depressed mothers (Stoneman & Berman, 1993). They were also less tolerant, less responsive and less nurturing toward their children and tended to generate guilt and anxiety in their children in order to control their behaviour (ibid). Moreover, parenting patterns related to depression were linked to antagonistic, hostile behaviour between siblings. Stress brought on by the demands of raising a child with a disability was found to adversely affect parental functioning and child rearing strategies, thus, interfering with sibling adjustment, and inherently, sibling relations (Crnic et al., 1983). In sum, parenting is a significant domain of FQOL as it represents the leadership and the cohesiveness of the whole family, hence its significance and relevance to this study.

Emotional Well-Being

Emotional well-being exists when *'the family has outside help available to take care of the special needs of all family members, and has the support they need to relieve stress. It means that family members have friends or others who provide support and have some spare time available to pursue individual interests .They have recurring life activities that help meet individual and collective needs'* (Turnbull et al., 2004, p.388-392).

There has been little research on the way in which children with disabilities can impact on their families' emotional well-being (Turnbull et al., 2004). Researchers found that parents of a child with disability devoted more time to caring for the child and spent less time in house work, especially when the child was younger. They also spent less time in social activities. In order to maintain a coordinated life, families changed work schedules, learned new childcare skills and redistributed house chores among family members (Gallimore et al., 1989). The emotional life of a family with a child with disability can vary according to the child's age, severity of disability and family's socioeconomic status (ibid).

The most discouraging finding of the study of FQOL in families with a child who has a disability was found in Canada, and related to *'the lack of practical support that families receive from relatives, friends, and their neighbours when dealing with their children's disabilities'*. One might expect that relatives would be more involved in practical support, but the families in this study indicated that this was not the case. Family and friends were more willing, however, to give emotional support to the families interviewed. It appears to be easier for others to provide emotional support for families with children with disabilities than it is to provide practical support. This is perhaps due to the lack of knowledge or comfort level that other family members or friends may have in working directly with children with disabilities, or, in some cases, it relates to geographical or other physical reasons that impede offers of support. Family satisfaction in this area was rated fairly low, as was initiative taken by families to find support. It appears that parents who have children with disabilities do not feel comfortable

enough to ask for help, or they assume that by asking, they will become a burden to others (Brown et al., 2003).

Previous research has demonstrated significant gender differences in perceptions of stress and reactive coping mechanisms. The socialisation hypothesis suggests that men are socialised to use instrumental coping strategies to deal with the perception of stress such as planning and executing strategies of action to deal with the perception of stress. Women, on the other hand, are socialised to use emotion-focused coping strategies, such as seeking the advice and comfort of others to deal with the presence of stress (Levy-Shiff, 1999).

For some parents, the birth or presence of a child with a disability may adversely affect pre-existing relationships with friends or relatives (McAllister et al., 1973). In one study, families with a severely child with a disability were found to be significantly less likely to participate in social activities or visit with friends, relatives and neighbours than parents without such a child (ibid). In another study, neighbours of families with a child with a disability were reported to be more 'standoffish', and tended to only interact with, or offer assistance to non-disabled siblings in the family (McAndrews, 1976).

Although the significance of social support has been recognised in families with children experiencing an condition or particular handicap (Kazak & Marvin, 1984; Marcenko & Meyers, 1991), few studies have extended those findings to explore the use of social support as beneficial or harmful to mothers of children with ADHD (Polodski & Nigg, 2001). Despite a general lack of research on the subject, there is some indication that mothers of children with ADHD has lack coping resources, including social supports. According to an early study, mothers of children with ADHD participate in social activities less often and report significantly higher levels of social isolation than mothers of children without ADHD (Mash & Johnston, 1983). Mothers of children with ADHD have also been found to have less contact with their extended family (Malacrida, 2003) and perceive supportive attempts made by family members as less helpful (Cunningham & Barkley, 1979).

In sum, emotional wellbeing as a domain of FQOL can be improved by the support element, which when provided, helps in decreasing stress and anxiety related to raising a child with TS, hence its significance and relevance to this study.

Physical / Material Well-Being

Physical / material well-being exists when *'the family receives medical and dental care when needed. Family members feel safe at home, work, school, and in their neighbourhood. Family members have transportation to travel wherever they need to, and have a way to cover their expenses'* (Turnbull et al., 2004 p.388-392).

Material well-being means being able to pay, at the very least for what the family needs and in some cases, for what it wants. Financial resources are needed for basic needs, health care costs, educational costs, transportation, financial assistance and extras. Researchers found that poor parents exhibit less positive behaviour such as hugs, praise or supportive statements toward their children (Lempers & Jacques, 1997), and often fight over money. Poor families showed less sensitivity and satisfaction with parenting, and higher income was related to higher marital satisfaction for fathers, and better child care (Willoughby & Glidden, 1995). Economic pressure was associated with lower marital quality and higher depression. Higher conflict and psychological distress was associated with frequent use of harsh discipline techniques and lower psychological functioning in children (Robila & Krishnakumar, 2006). Children of poor families are more often excluded from high quality childcare environments, better schools, safer play areas and positive adult role models (Park & Turnbull, 2002).

The cost of increased medical expenses, equipment, therapy and specialised child care associated with a child with a disability places an additional strain on families' financial resources (Turnbull & Turnbull, 1990). Crnic et al., (1983) reported that families of children with disabilities experienced reduced social mobility, and consequently, limited financial advancement. Mothers of children with disabilities often gave up their careers or worked only part-time in order to meet the needs of their children

with disabilities. In sum, financial wellbeing as a domain that determines FQOL is subjective and relative to the family's perception of their ability to cater for their needs in coping with having a child with TS, hence its relevance to this study.

Support for the Child with TS

Support for a family member with a disability exists when *'the family member with special needs has support to make progress at school, at work, and at home. It means that the family member with special needs has support to make friends, and has a good relationship with the service providers who work with him'* (Turnbull et al., 2004, p.388-392). The services that are available for families who have a child with a disability are not always appropriate or do not always meet the needs of the families. The needs of these families are constantly changing, therefore, a community agency that is helpful one day, may not be so useful at another time, depending upon the family's needs at that point in time (Browne et al., 1997).

The domain of FQOL – 'Disability-Related Support' – focuses on inner-family and extra-family support provided in order to benefit the family member with a disability (Turnbull et al., 2004). Sample items include:

1. My family member with special needs has supports to achieve goals at school or in the work place.
2. My family member with special needs has supports to achieve goals at home.
3. My family member with special needs has supports to make friends.

Several studies have investigated the various ways in which social support influences the family. In particular, the benefits of social support have been associated with positive family adjustment, greater maternal satisfaction, lower maternal stress (Kazak & Marvin, 1984), and reduced marital and parent-child conflict in families of children experiencing a condition or 'handicap' (Marcenko & Meyers, 1991).

Yet, some researchers have also suggested that social support can be potentially harmful and cause even greater distress (Harris, 1992). Supportive attempts can sometimes be perceived as intrusive and officious (Aneshensel et al., 1993). Additionally, increased levels of stress and marital discord have also been associated with higher rates of family involvement in personal affairs (Kazak & Marvin, 1984).

Most researchers have failed to formulate a clear and precise definition of social support which can be demonstrated by valid and reliable indicators (Thoits, 1981). The various dimensions and functions of social support have been uniquely evaluated across studies (Lin et al., 1986). Since the concept of social support is often conceptualised and measured in different ways, it is extremely difficult to make legitimate comparisons between the different studies. Therefore, the specific attributes or features of social support which function to reduce the impact of stress cannot be accurately identified (Thoits, 1981).

Previous research has demonstrated significant gender differences in perceptions of stress and reactive coping mechanisms. The socialisation hypothesis suggests that men are socialised to use instrumental coping strategies to deal with the perception of stress such as planning and executing strategies of action to deal with the perception of stress. Women, on the other hand, are socialised to use emotion-focused coping strategies, such as seeking the advice and comfort of others to deal with the presence of stress (Levy-Shiff, 1999). Support as a domain of FQOL is related to the feeling of social solidarity, counselling, and sharing of difficulties which is significant in the case of families having a child with TS, hence its relevance to this study. This section has discussed the aspects of FQOL. The next section discusses the systems theory, which was chosen to underpin the study.

2.11 Systems Theory

Systems Theory was chosen to provide one of the theoretical foundations for the research since it enabled a conceptualisation of the family as a system, facilitating the clarification of the dynamics of the various elements involved in the family.

Since the 1940s attempts have been made to construct a theory that would address the manner in which systems are constructed and behave. Social workers and other care-giving professionals of various disciplines have adopted the general principles of theoretical models that may be applied in research, such as the 'General Systems Theory' developed by Ludwig von Bertalanffy (1968). Von Bertalanffy wanted to establish a theory that would guide research in a number of disciplines, as he saw remarkable parallels between them. His assumption was that if these disciplines focused their research and efforts into developing their own theory, they would identify common rules and principles that would be applicable in a number of types of systems. He argued that if there was a common framework for all systems, research findings concerning a particular field of study would be applicable and useful for researchers in other fields. He believed that with time, what would be discovered in these studies would be applicable in the real world in general.

After more than fifty years in investigating systems, Systems Theory has developed to the point where many of the concepts of this theory are commonly accepted in our daily language. Thus it is common practice to talk about the health system, the family system, body system, information system, banking system, political system, etc. One of the reasons for this categorisation is that the amount of knowledge and information regarding systems has increased significantly in the modern era. Since it is impossible to know everything there is to know about the world, methods are discovered to organise information, in order to avoid an overload of information. Thus, individuals and especially researchers focus on small areas of knowledge, and do not try to absorb everything at once.

Systems Theory is a trans-disciplinary theory that brings together theoretical principles and concepts from ontology, philosophy of science, physics, biology and engineering. Systems Theory was the name used by early investigators to describe organisation and interdependence of relationships (Rapoport & Rapoport, 1969), and was defined as a set of objects or elements in interaction to achieve a specific goal (Ryan & Gates, 2004). The function of any system in general is to convert or process energy, information, or materials into a product or outcome for use within the system, or outside of the system (the environment) or both (ibid). Since this study focuses on FQOL, systems theory may explain how members of families who have a child with TS perceive their quality of life.

Despite rich explanations for Systems Theory in the literature, most researchers do not clearly identify the hypotheses behind the theory. One of the basic assumptions of Systems Theory is 'holism', that is, that life phenomena stem from a single complete principle (Bateson, 1979).

Another assumption is that Systems Theory is organised hierarchically, that is, systems are nested inside other systems (Fisher et al., 1985). For example, family members would be the components of the family system, and the family would be a component in the neighbourhood system.

2.11.1 Family Systems Theory

Systems Theory, considers the way that relationships within the family and between the family and social environment influence individual development and family functioning, provides guiding principles for family systems (Bowen, 1978). The principles of family systems are based on family relationships, how families function and how families and communities interact. Therefore, Family Systems Theory (ibid), is based on an ecological model which views human growth and development in the context of its environment and relationships and which observes the interactions of humans with their environments (ibid; Kreppner & Lerner, 1989). From an ecological perspective, the most logical model of a family is as a system, (Hinde, 1989). Most researchers now approach the family from what could be called a 'systems perspective' (Kreppner & Lerner, 1989).

Family Systems Theory has had a significant impact on the study of families and on approaches to working with families. It has guided research in such areas as understanding traumatic events or chronic health issues and their impact on individuals and families (Bowen, 1978; Fingerman & Bermann, 2000). It has provided a useful lens through which a greater understanding of families has emerged. However, as with any other lens, critics have challenged the clarity of the lens in certain areas. Some critics have argued that issues of gender inequality are not fully articulated or addressed within Family Systems Theory. For example, in patriarchal societies, where power lies primarily with men, equality of influence between men and women cannot be assumed. Critics of Family Systems Theory argue that such inequality is often overlooked or understated (Goldner, 1989; Yllo, 1993).

Over the years variants in Family Systems Theory have emerged. The Communications Model focuses on the communication patterns found within family systems, specifically on the role of inputs and outputs in communication and the consistency between these in explaining family communication patterns in functional and dysfunctional families (Watzlawick et al., 1967). In contrast, Salvador Minuchin's (1974) work with Family Systems Theory has focused more on the spatial nature of families. Central to this orientation is an examination of the social contexts and structures in which families find themselves and their interaction with those contexts and structures. A Family Systems framework has developed to guide professionals in gaining an individualised perspective of each family's strengths and needs for family support (Turnbull & Turnbull, 2001).

The ecological systems approach has been used in research to study families of children with ADHD. A few research studies attempted to employ a family systems approach in research concerning families of children with ADHD (Bernier & Siegel, 1994). They found evidence of dysfunction in the family. In these cases, the ecological systems approach was applied to examine the social context and other relevant factors, aside from aspects of the individual child's disorder as factors which might contribute to this dysfunction (Bernier & Siegel, 1994).

The ecological systems approach to the study of the family maintains that a change or difficulty experienced by one family member ultimately impacts the entire family unit and creates the need for family adjustment or adaptation (ibid). Research indicates that the behavioural characteristics of children with ADHD cause considerable disruption to the family environment and the psychological functioning of the family; especially the mothers (ibid).

Maternal stress and family strain has also been associated with increased behavioural problems in children. Thus, the mother-child relationship in families of children with behavioural problems is highly susceptible to cyclical patterns of dysfunctional behaviour (ibid). Yet, as noted above with respect to FQOL, little is known about how families deal with the added stress associated with raising a child with TS. An ecological systems approach encourages consideration of the social context and other relevant factors apart from aspects of the individual child's disorder which may contribute to this dysfunction. Therefore, this research considers how the FQOL of families with children with TS is affected not only by the behavioural problems of their child but also by other variables such as neighbours, friends, the educational system, and the psychological and medical services provided to the child with TS.

The usefulness of ecological model and family systems approach has frequently been illustrated in the study of families with a child with disability. Results of such studies indicate that the presence of a child with disability places intense stress on the family and its total resources, including physical, psychological, and financial resources, as well as its social supports (Bubolz & Whiren, 1984; Birnbaum et al., 1997). It was found that other matters, such as poverty, unemployment, defaulting child-support payments and inadequate schools, administrators or teachers can exacerbate family stress (Bernier, 1999). Researchers speculate that if sufficient resources are not accessible to families with a child with disability, family adaptation may not be possible. *'The physical, social and emotional functioning of family members is profoundly interdependent, with changes in one part of the system reverberating in other parts of the system'* (McGoldrick & Gerson, 1985, p.1).

Therefore in order to understand the influence of the child with TS living at home on the FQOL, Systems Theory and the ecological model seemed to the researcher to provide the most logical explanatory theory.

2.11.2 The applicability of the Ecological Paradigm to this study

The ecological paradigm is drawn from psychology and sociology focusing respectively on child-father-mother-sibling interactions, and on marital relationships and the family as a whole in a social context. *'When we examine the family from an ecological point of view, no one person or thing can be realistically identified as the "cause" of a problem'* (Yerby *et al.*, 1990, p.63).

An ecological perspective views human development from a person-in-environment context, and emphasises the interconnections between the organism and the environment (Kreppner & Lerner, 1989). Accepting the principle that all growth and development take place within the context of relationships means that a child must be studied in the context of the family environment and the family must be understood within the context of its community and the larger society. Although parents have a profound influence on the ability of the child to develop in a healthy, competent manner, children also influence their parent's behaviours. Therefore, when dealing with a child's behaviour, or addressing a family's financial, emotional and social needs, it is necessary to consider not only the individual but also the contributing factors from the environment and the interpersonal relationships. In this context, the 'goodness of fit' model which refers to the match between the developmental needs of children and the demands, resources and capacities of their family, school and community environments, can be useful in indicating what is needed to support and strengthen families (Lerner, 1993).

Additionally, because families develop, they must respond to the demands and expectations of their environment such as work, social groups, community institutions and the society as a whole. Stress builds when the resources and coping skills of a family are inadequate to meet the demands and expectations of the social environment.

Family stress levels are a predictor of negative outcomes for children. If stress increases beyond a certain point, for whatever reason, a family's ability to nurture its children decreases (Schorr, 1989). The interactions within and between the different environments of a family make up the 'ecology' of the family and are key elements of an ecological perspective. The environments of a family's ecology include:

Family: The family performs many functions for its members essential to healthy development and mediates between the parents and siblings environments.

Informal Social Network: A family's social network grows out of interactions with people in different settings; extended family, social groups, recreation, and work. Ideally, this network of caring for others shores up feelings of self-worth, mobilises coping and adapting strategies, and provides feedback and validation.

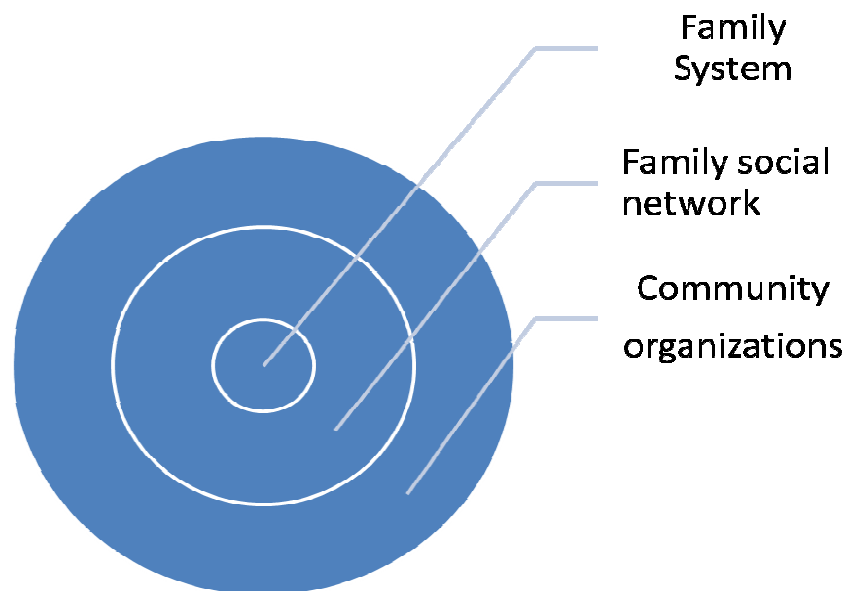
Community Professionals and Organisations: A community's formal support organisations provide families with resources related to professional expertise and/or technology.

Society: Social policy, culture, the economy define elements of the larger ecology that impact the way a family functions.

Bronfenbrenner's Ecological Systems theory (1979) considers the child's development within the context of the system of relationships that form their environment. Bronfenbrenner's theory (ibid) relates to different 'layers' of environment, each having an effect on a child's development. This theory has recently been renamed '*Bioecological Systems Theory*' indicating that a child's own biology is a primary environment fuelling their development. The interaction between factors in the child's maturing biology, the immediate family/community environment, and the society influences the child's development. Changes or conflict in any one layer affect the other layers. This means that any investigation of a child's development must not only understand the child and the immediate environment, but also relate to the child's interaction with the larger environment.

Figure 1 below depicts the ecological system framework of Bronfenbrenner including factors affecting the FQOL.

Figure 1: Bronfenbrenner's Ecological Systems Framework



In the present research, the family system is composed of the father, mother, brothers and sisters, and the child with TS. According to Family Systems theory, FQOL is affected by a process of inputs and outputs. The *inputs* are the external and internal factors. The internal factors are the family's income, socio-economic status of the family, parents' age, brothers and sisters, parents' level of education, age of the child with TS, and the child's degree of disability. The external factors are extended family, neighbours, friends, the educational system, and the psychological and medical services provided to the child with TS. The outputs that are affected by the family inputs are the family's perceptions of the importance of different domains of FQOL and their levels of satisfaction in these domains (Bowen1978; Park et al 2003; Turnbull et al., 2004; Murray & Lampinen, 2006).

2.12 Summary

The present research aims to examine the FQOL of families of children with TS. Since there is a dearth of theoretical knowledge regarding FQOL, knowledge regarding individual QOL was used to help construct a framework that would explain and outline FQOL. A focus was directed to five FQOL domains: family interaction, parenting, emotional well-being, financial well-being, and support for the family member with a disability, which are the main domains studied in the present research. Family Systems Theory and the Ecological Systems framework were reviewed as the most appropriate base for the methodology of this research, and to delineate the theoretical background.

CHAPTER 3: THE CONCEPTUAL FRAMEWORK OF THIS RESEARCH

3.1 Introduction

Conceptual frameworks provide a theoretical overview of the intended research and create order within the research process (Trafford, 2007). Additionally, Bell (2005) maintains that the conceptual framework provides the process by which the data will be collected, analysed, and interpreted. Moreover, Miles and Huberman (1994) suggest that a conceptual framework is a visual or written product, one that explains the main issues under study, namely, the key factors, concepts, or variables – and the possible relationships among them. This chapter presents the conceptual framework of this research, the links between the concepts included in the framework and the considerations for selecting them, as a foundation for the investigation of the FQOL of families who have a child with TS.

3.2 Constructing the Conceptual Framework

The conceptual framework of this research draws on theories of psychology, because the study deals with FQOL including the emotional welfare of families who have a child with TS, sociology, because the concept of a family lies at the core of this examination, and psychiatry, relating to the TS diagnosis and treatment.

More specifically, this research sought to fill the gap in knowledge regarding the FQOL of families with a child with TS, by studying and understanding the perceptions of the family members. Thus, the research addressed the following research questions:

1. Which FQOL domains are perceived by family members to be affected by the fact that they have a child with TS?
2. How do different families raising a child with TS perceive their FQOL?

Research and theoretical literature relates extensively to TS (Dreher, 1996; Abelson et al., 2005; Swain et al., 2007), the ways to diagnose children with TS (Dreher, 1996; Pauls et al., 1993; Merck Manual of Medical Information, 1997), and how to treat them (ibid). More specifically, the literature offers enough knowledge regarding the medical (ibid) as well as the social aspects (Sukhodolsky et al., 2003; Bawden et al., 1998; Carter et al., 2000) of children with TS. There also appears to be a significant body of literature concerning quality of life of children with TS (Browne et al., 1997; Turnbull et al., 2000; Elstner et al., 2001; Goodman et al., 2006), but the researcher, to date, has not found any literature regarding FQOL of families with children who have TS.

The complexity of the behaviours presented by children with TS, for example: vocal, facial, and body tics, with severe behavioural and learning difficulties, combined with OCD and ADHD, may lead to internal and external responses of discomfort for people who come in contact with those children (Greene, 1996). With no clear idea of what this disorder entails, families often lack necessary knowledge of how to manage their children. As

a result, the daily difficulty for families with children with TS, at home, in the educational frameworks, and in various social frameworks, may cause a feeling of frustration among family members, which consequently might affect the entire FQOL.

Research that would reveal perceptions of parents and siblings of a child with TS regarding their FQOL could increase the understanding of what this syndrome entails and guide policy makers and care givers as to how to maximise treatment and how to design intervention programmes to improve practice.

Since the research sought to examine how families who have a child with TS, perceive their FQOL, and therefore needed to relate to the family as a unit or system, it was decided that the research would be based upon systems theory and an ecological model.

Therefore, the following concepts were selected as the components of the conceptual framework that underpinned the research:

FQOL domains were selected as the focus of this study. QOL has been defined as the difference between the hopes and expectations of the individual and their present experience (Calman, 1984), and is experienced when a person's basic needs are met and when he or she has the opportunity to pursue and achieve goals in major life settings, namely, work, school, home, and community, (Goode, 1988). Therefore, in this research the family members' perceptions of the importance of those domains and their level of satisfaction in these domains were measured, in order to assess the FQOL. A systemic approach to the family was adopted in an attempt to explain the processes of interaction between its members. In the context of this research, the family is defined as: A kinship group comprising one or more children diagnosed as having TS, together with two parents or single parent and siblings living in one household.

Systems Theory and the ***Ecological Model*** were selected to explain how the family operates and functions as a system, with a child with TS. Systems Theory was the name used by early investigators to describe organisation and interdependence of relationships (Rapoport & Rapoport, 1969), and was defined as a set of objects or elements in interaction to achieve a specific goal (Ryan & Gates, 2004). Systems theory considers the way relationships within the family and between the family and social environment influence individual development and family functioning, and provides guiding principles for family systems (Bowen, 1978). Thus, Systems Theory explains how family members interact and what personal and interpersonal processes occur inside the family.

The Ecological Model views human development from a person-in-environment context, and emphasises the reciprocal connections between the person and the environment (Kreppner & Lerner, 1989). Thus, the ecological model delineates how family members interact with the environment outside the family. Using the two theories, (Systems Theory and the Ecological Model) enables a holistic depiction of the family functioning inside and outside the family system. Figure 2 below presents a visual representation of the conceptual framework of this research.

Figure 2: Graphic Representation of the Interactive Process between the Ecological Model and Systems Theory

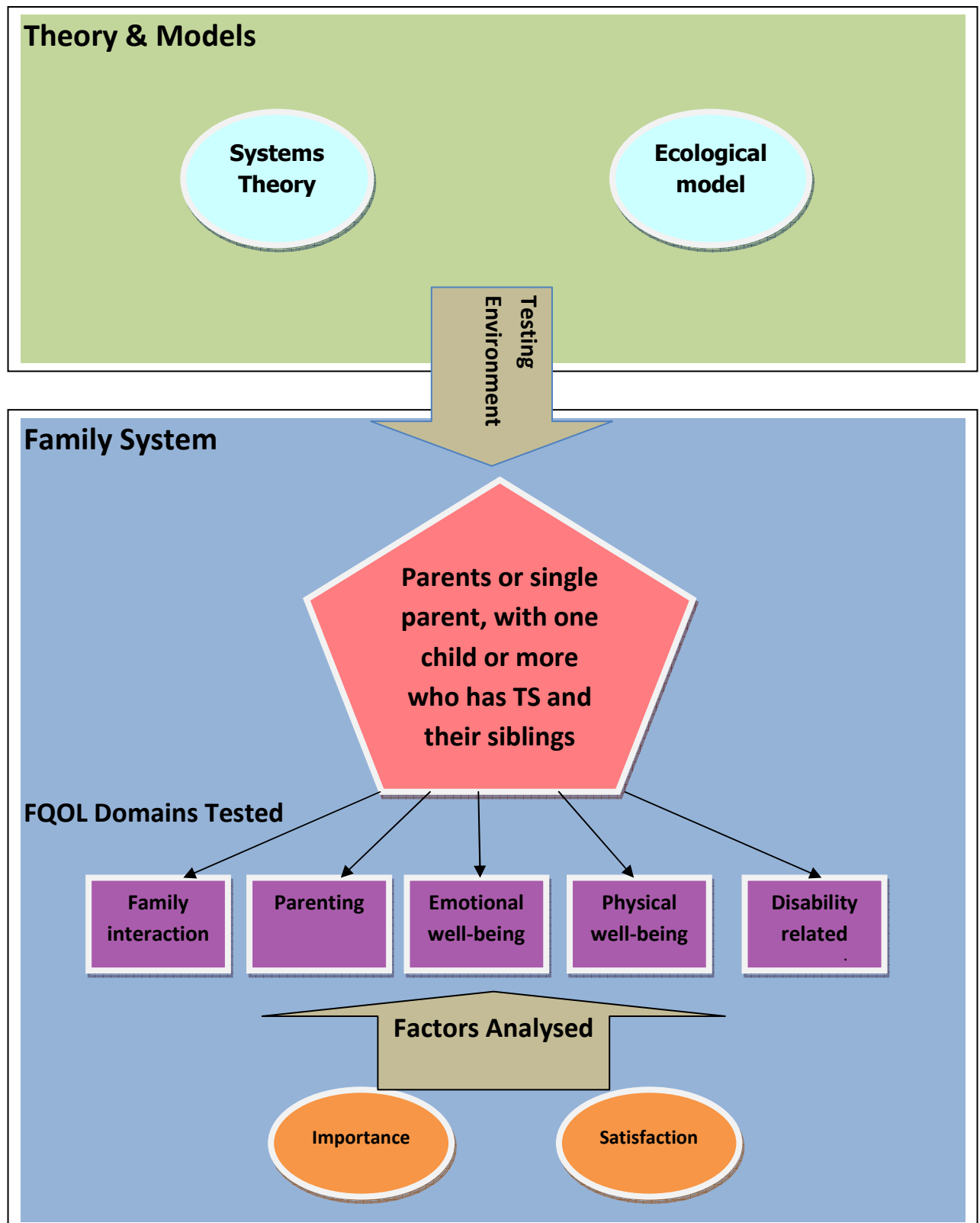


Figure 2 above shows the interaction of the components that were involved in the research process and the way that they complement each other. Systems Theory and the Ecological Model are used in this research to explain FQOL. The family system is composed of parents or a single parent, siblings and the child with TS. The family perceptions of the importance of the different domains of FQOL and their levels of satisfaction in these domains are the indicators of their FQOL. According to the model, the family system is located in the centre of the model, in order to demonstrate the centrality of the family unit and the ways in which Systems Theory and the Ecological Model explain the dynamics inside and outside the family that has a child with TS. Thus, the child with TS as part of the family affects the type of parenting and the relationships among the siblings. Similarly, the child with TS affects the dynamics between the family and the environment outside the family unit. The dynamics inside and outside the family unit generates the FQOL as an outcome.

Ultimately, the research advanced an understanding of the perceived FQOL of families who have a child with TS within the Israeli context that was presented in this research as a case that could have implications for better treatment practice and training of treatment providers in this field.

The goals of this research were achieved through the application of a mixed methods research, combining quantitative research with qualitative research traditions. The next chapter presents the methodological considerations that underpinned the choice of methods and tools used in this study.

CHAPTER 4: METHODOLOGY

4.1 Introduction

This chapter first states the main research goal, the research questions, and the research boundaries. It elucidates the choice of the research approach appropriate for the conceptual framework of this research and clarifies considerations involved in constructing the research design, explaining why and how the mixed-methods approach and the different research methods were chosen, relating to each method according to the order of these methods in the research design.

Additional sections of this chapter relate to the issues of validity, reliability, and generalisability. A critical depiction of the sampling process and selection of the research population used in this research is provided. The ethical considerations that ensured the privacy and the safety of the respondents are explained. An additional section describes the researcher's role and level of involvement in the data-collection.

Finally the data analysis methods are presented relating to the decision to reinforce the validity of the research data with the strategy of triangulation, so that this chapter presents the background for the analysis of the findings presented in Chapter 5.

4.2 The Research Goal and the Research Boundaries

The research aimed to fill a gap in knowledge concerning the FQOL of families who have a child with TS living with them in the family home. The study therefore excluded families whose child with TS normally resided outside the family home. The research focused on the nuclear family of the child with TS and so no attempt was made to relate to other extended family members even if they resided in the same home with the nuclear family.

4.3 The Research Questions

The research seeks to address two main questions with eleven sub questions:

1. Which FQOL domains are perceived by family members to be affected by the fact that they have a child with TS?

- A. Are there differences between the perceptions of mothers and those of fathers regarding the importance of the FQOL domains and their level of satisfaction in those domains?
- B. What are the perceptions of father, mother and the family as a whole, regarding the services they perceive as necessary for the child (ren) with TS and the services which are actually received for the child with TS?
- C. What are the perceptions of the father, mother and the whole family, regarding the services they perceive as necessary for the family and the services which are actually received for the family?
- D. What are the differences between the perceptions of fathers and perceptions of mothers regarding the services they perceive as necessary for the child with TS and the services which are actually received for the child with TS?
- E. What are the differences between the perceptions of their FQOL by parents of the child with TS and those of the child's siblings?
- F. What correlations can be found between the fathers' perceptions of the FQOL Domains and their perceptions of Services for the Child and Family?
- G. What correlations can be found between the mothers' perceptions of the FQOL Domains and their perceptions of Services for the Child and Family?
- H. What correlations can be found between the perceptions of both parents' regarding the FQOL Domains and their perceptions of Services for the Child and Family?

2. How do different families raising a child with TS perceive their FQOL?

- A. Do families of children with TS who perceive their child as having a severe form of TS have a different perception of their FQOL than families who perceive their child as having a mild form of TS?
- B. Do families of children with what is perceived as a severe case of TS have different perceptions of the services variables than families of children with what is perceived as a mild case of TS?
- C. Is there a difference between families of children with what is perceived as a severe case of TS and families of children with what is perceived as a mild case of TS in the indices of difference of services for the child?

4.4 Choosing an Appropriate Research Approach

Arguably, there are two main approaches to social research: the classic 'scientific' positivist, quantitative approach and the qualitative approach. A positivist quantitative research approach is typically based on statistics generated through the use of large-scale surveys, using methods such as questionnaires or structured interviews. This type of research can reach many more people, but the contact with those people is much briefer than it is in qualitative research. Measurement is often regarded as being only a means by which observations are expressed numerically in order to investigate causal relations or associations. Creswell (1994) defines quantitative research as a type of research *'explaining phenomena by collecting numerical data that are analyzed using mathematically based methods (in particular statistics).'* Thus, in the social sciences, quantitative research refers to systematic empirical investigation and measurements of different phenomena by their quantitative properties and the relationships between them.

In contrast, qualitative research makes an in-depth investigation of research subjects exploring, behaviour and experiences through such methods as semi-structured and unstructured interviews or focus groups. Because of the desire for greater depth and 'rich' descriptions, fewer people take part in the research, but the contact with these people tends to last a lot longer. Cronbach (1975) states that *'the special task of the social scientist in each generation is to pin down the contemporary facts. Beyond that, he shares with the humanistic scholar and the artist in the effort to gain insight into contemporary relationships'* (p. 126).

Qualitative research, broadly defined, means *"any kind of research that produces findings not arrived at by means of statistical procedures or other means of quantification"* (Strauss and Corbin, 1990, p. 17). Where quantitative researchers seek causal determination, prediction, and generalisation of findings, qualitative researchers seek instead illumination, understanding, and extrapolation to similar situations.

Qualitative research is often used to gain a general sense of phenomena and to form theories that can be tested using further quantitative research, to gain better understanding of such things as a person's intention (from the speech response of the participant) and meaning .

4.4.1 Optimising the Research Approach

Over the years there has been a large amount of complex discussion and debate surrounding the topic of research methodology and the theory of how inquiry should proceed. Much of this debate has centred on the issue of qualitative versus quantitative inquiry – which might be more ‘scientific’. Different methodologies become popular at different social, political, historical and cultural times in our development, and, in my opinion, all methodologies have their specific strengths and weaknesses. Because typically qualitative data involve words and quantitative data involve numbers, there are some researchers who feel that one is better (or more scientific) than the other.

The researcher may employ either inductive or deductive thinking or process to investigate the chosen phenomenon. Inductive research is based on inductive reasoning which transforms specific observations into general theory, a thinking process moving from the specific to the general. Observing a pattern in society, the researcher may form a hypothesis or assumption on it, and then conduct surveys or experiments to verify the hypothesis and reach a conclusion. In other words a qualitative researcher using inductive thought *'begins with a research question and little else. [The researcher] begin(s) with detailed observations of the world and move(s) towards more abstract generalisations and ideas'* (Neuman, 1997:334). While in the deductive tradition the researcher employs deductive thought which transforms general theory into specific hypothesis suitable for testing, starting *'with an abstract, logical relationship among concepts then move(s) towards concrete empirical evidence'* (ibid: 46). The researcher therefore formulates a particular theoretical framework and then sets about testing it. In this case the researcher's thinking runs from the general to the specific.

Although qualitative research usually employs an inductive process, nevertheless a hypothesis is not needed to begin research. In contrast, all quantitative research requires a hypothesis before research can begin and usually employs deductive thinking. However quantitative research can certainly be inductive i.e. making numerous observations without a prior hypothesis and, arriving at conclusions as a result of quantitative analysis of these observations.

Some researchers believe that qualitative and quantitative research can be effectively combined in the same research project (Strauss and Corbin, 1990; Patton, 2002). For example, Russek and Weinberg (1993) claim the use of both quantitative and qualitative data enabled them to gain insights that neither type of analysis could provide alone. Since the present research sought to examine the FQOL of families who have a child with TS, and with no pre-determined hypotheses available, the study adopted an inductive research design approach (Creswell, 2003), yet this was combined with mixed methods research.

Based on the development of both quantitative and qualitative research methodology, researchers in social and human sciences have adopted mixed methods approaches, which employ strategies to collect and analyse both qualitative and quantitative data (Creswell, 2003). Mixed-methods approaches enable the researcher to conduct method triangulation in order to discover convergence across qualitative and quantitative methods (Patton, 2002). A mixed methods approach is expected to create a more reliable explanation through triangulation. In this research, a mixed methods approach was adopted, applying the different principles of each method in order to reinforce the research data. The present study therefore implemented mixed-methods research strategy, (Johnson & Christensen, 2004; Creswell, 2003; Ortiz & Greene, 2007) in order to gain a holistic view of this phenomenon

A mixed-methods approach integrating different methods is likely to produce better results in terms of quality. Mixed-method strategy is a way to produce creative alternatives to traditional or more monolithic ways to conceive and

implement evaluation. These alternatives will not be able to make radical shifts in the short run. However, they can lead to a more reflexive and more critical approach to the evaluation practice and, ideally, be more useful and accountable to broader audiences (Caracelli & Greene, 1997).

The present research was a 'mixed methods' research based on a qualitative research approach but with a limited use of the quantitative approach to obtain some initial data. It therefore combined the use of both quantitative and qualitative research instruments (Creswell, 2003; Curry et al., 2009). This strategy allowed the researcher to gain a broader and more precise picture of the phenomenon under investigation.

It is noted that the quantitative data was gathered at the initial stage of the research. Critical views of quantitative research (Bryman, 2004) posit that the size of the sample from which data is derived is likely to impose limitations on the sort of analysis conducted and on the confidence assigned to the findings. In this research data were obtained from 25 families having a child with Tourette syndrome. Additionally, in each family, at least four members responded to the closed-ended questionnaire. Therefore, data were collected from 100 respondents, which make an appropriate body of data to be analysed statistically.

Qualitative description enabled the researcher to draw conclusions and create general rules that were based on the quantitative results. This allowed the research questions to be answered by describing a correlation between variables. Although quantitative research may evolve from a clear and structured theory, and the research design is predetermined, data collection from questionnaires and statistical data analysis were judged to be insufficient for the present research, because the research examined FQOL i.e. family systems and interactions between individuals in the family and between individuals and the family systems. Quantitative research cannot provide in-depth responses to these issues.

Similarly, a qualitative research method used alone would produce comprehensive personal information, but it would be difficult to generalise this information to other families (Mason, 2006). Use of qualitative research provides data for the investigation, description and explanation of family members' behaviour and perceptions from their own viewpoints. This research method allowed the researcher to develop subjective understanding, without any hypotheses, producing interpretive descriptions of the feelings, thoughts and perceptions of the participants. Qualitative research provided valuable information through an in-depth understanding of the way that the respondents understood and interpreted their world. This enabled the researcher to map the world of content relevant to the research and to map deeper layers of human perception, and improve understanding of how people perceive their world, providing deep psychological insights that quantitative research could not penetrate, but this method could not be used exclusively. Exclusive use of this method would prevent replication of the research, as it would be more subjective and restricted to a particular time and place. By nature, it is also less focused on attempts to investigate causality.

For these reasons, although Interviews were used to gather qualitative data that would explain the feelings of families and to understand the depth of the impact of the existence of a child with TS on the family; a questionnaire was also used to examine the families' FQOL since the responses to this questionnaire could provide quantitative information regarding various domains of FQOL.

A note needs to be taken here regarding the quantitative data that was gathered at the initial stage of the research. Critical views of a quantitative research (Bryman, 2004) posit that the size of the sample from which data is derived is likely to impose limitations on the sort of analysis conducted and on the confidence assigned to the findings. However, in this research data were obtained from 25 families having a child with Tourette syndrome. Additionally, in each family, at least four members responded to the closed-ended questionnaire. Therefore, data were collected from 100 respondents, which make an appropriate body of data to be analysed statistically.

This strategy facilitated the performance of triangulation, which was expressed in the present research through the comparison of data from two types of instruments and data relating to the different levels within the family (mother, father and siblings). Triangulation reinforced the research reliability and enabled generalisation of the research results in a significant manner. Figure 3 below illustrates the triangulation employed in this study.

Figure 3: The Research Triangulation

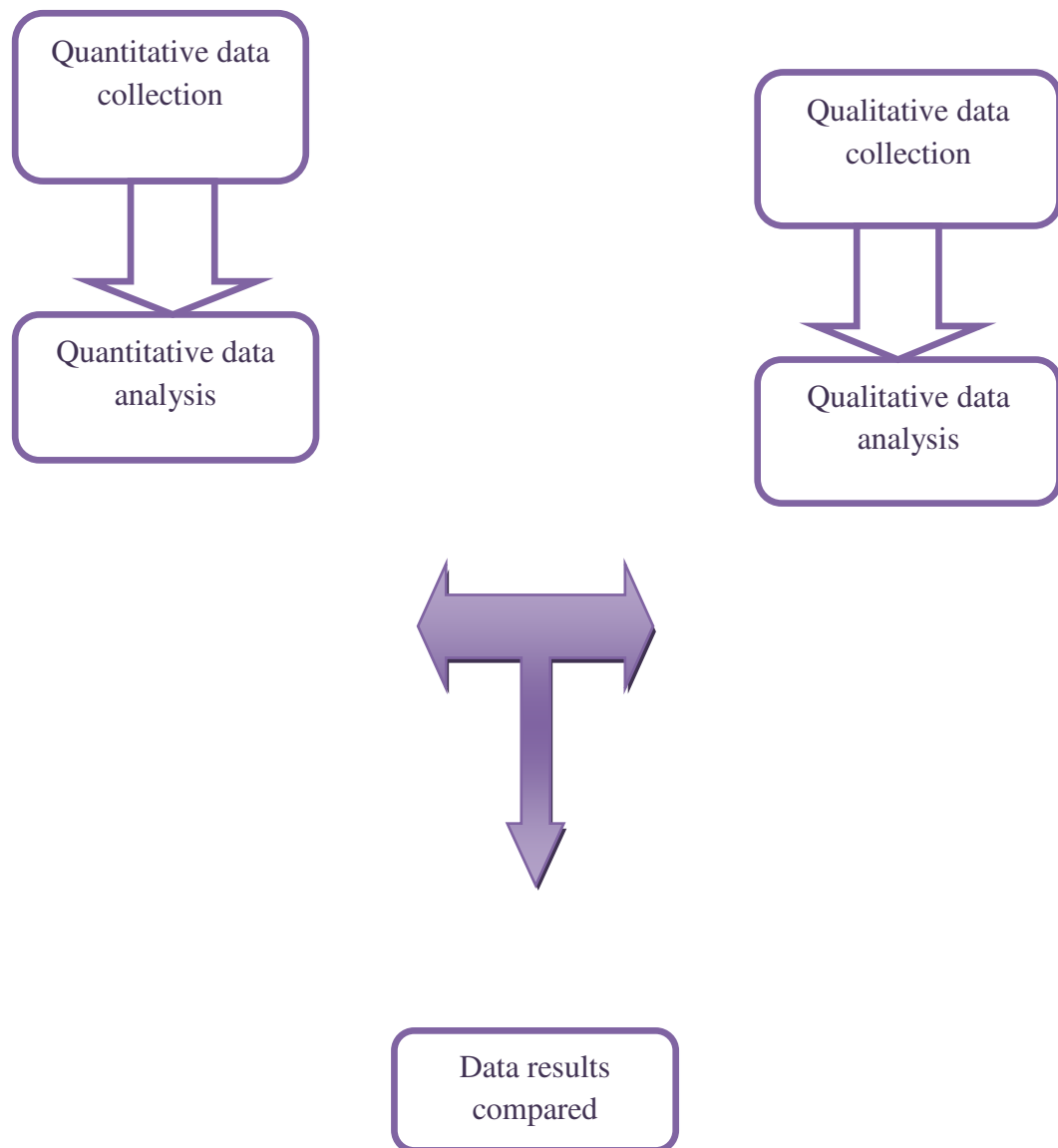


Figure 3 shows that in Stage 1 data were gathered using questionnaires for the fathers, mothers and siblings, and then analysed statistically. In Stage 2 data were gathered using interviews with parents of children with TS, and these data underwent contextual/ OR textual/ OR narrative analysis. Stage 3 of the study compared the results that emerged from the questionnaires with those that emerged from the interviews.

4.5 Research Techniques and Instruments

As noted above, the present research was based on a mixed-methods research approach (Greene & Caracelli, 1997; Burke Johnson & Onwuegbuzie, 2004) including two methods of data collection, derived from the quantitative and qualitative approaches, utilising the instruments of a closed-ended questionnaire and an unstructured interview.

4.5.1 The Closed-Ended Questionnaire

The research questionnaire that was adopted in this research, entitled the *Beach Centre Quality of Life Scale* was developed by Park et al. (2003) and Turnbull et al. (2004) to examine FQOL. Schalock et al. (2002) suggested that this FQOL scale is the only acceptable means of determining outcome of services and policies. Researchers in Canada and the United States provided a framework for ascertaining perceived quality of life in families. Canadian researchers developed a theoretical framework and an accompanying survey for gathering information about FQOL using the Beach centre scale (Brown et al., 2003).

As explained in Chapter 2: Theoretical Perspectives, on the basis of extensive qualitative (Poston et al., 2003) and quantitative (Brown et al., 2003) analyses, investigators of FQOL identified five specific domains of FQOL. These findings were the basis for a pilot version of a Beach Centre FQOL Scale. Park et al. (2003) conducted initial development and psychometric evaluation of the pilot scale. Cronbach's α for the total instrument is .94 (Summers et al., 2005). Each of the five subscales displays unidimensionality and internal consistency: Family Interaction ($\alpha=.92$), Parenting ($\alpha=.88$), Emotional Well-being ($\alpha=.80$), Physical/Material Well-being ($\alpha=.88$), and Disability-Related Support ($\alpha=.92$).

This scale measures levels of family members' perceptions of the importance of different domains of FQOL and their levels of satisfaction with regard to these domains. The scale contains twenty five items and five subscales:

- Family Interaction (6 Statements)
- Parenting (6 Statements)
- Emotional Well-Being (4 Statements)
- Physical / Material Well-being (5 Statements)
- Disability-Related Support (4 Statements)

The Beach Centre FQOL Scale constitutes a good starting point from which to address impact evaluation, as it is the only available quantitative tool on FQOL (Schalock, 2000). In addition to this FQOL scale, the questionnaire employed in the present research included three additional sections:

1. Statements regarding support and services for the child with TS (11 statements).
2. Statements regarding support and services for the family (14 statements)
3. Requests for general family data and personal details of the respondent.

The salient feature of the FQOL scale is that results are mapped so that the dimensions of importance and satisfaction are compared (ibid; Schalock et al., 2002). This enables users to give particular indicators by choosing one of the following quadrants:

1. High importance/Low satisfaction (priority indicators for more support).
2. High importance/High satisfaction (indicators representing family strengths).
3. Low importance/Low satisfaction (indicators that are not priority areas for more support).
4. Low importance/High satisfaction (indicators that are not priority areas for more support).

In addition, statements regarding support and services for the child with TS and services for the family were also mapped so that the relations between the necessity of services and received services are compared in accordance with the dimensions of importance and satisfaction.

The research questionnaire was distributed to parents of children with TS, and to the siblings of children with TS. The questionnaire for the siblings included only two sections: Statements regarding FQOL, and Personal details of the respondent.

The original questionnaire had to be translated into Hebrew as it was originally written in English. To minimise bias, and to ensure validity and reliability, the translation was performed by a professional translator. The questionnaire was then examined and modified by an expert in the Hebrew language. It is noted that the questionnaire was devised within one cultural context, and adapted to fit the Israeli cultural context through the translation.

In order to assure the validity of the questionnaire for the Israeli context, the researcher then piloted the questionnaire with parents from five families without children who had been diagnosed with disabilities, and asked them to report any problems in understanding, any technical problems in answering, how long it took to fill in the questionnaire, etc. It was felt justifiable to administer the pilot questionnaire to parents of children without disabilities since the focus was on the accuracy of translation rather than on the substantive issues of the research. After receiving and relating to their comments, the questionnaire was adjusted. Given the results of the pilot, and since the questionnaire aimed to reveal perceptions of FQOL of families who had children with TS, the questionnaire seemed appropriate to be used within the Israeli context.

4.5.2 Interviews

Interviews are conventionally used to collect information about opinions, perspectives and meanings, and to obtain interviewees' insights. Interviewers seek to understand central themes in the lives of research interviewees and the meanings that they give to these themes, on both

factual and conceptual levels. The main task in interviewing is to understand the meaning of what the interviewees say (Kvale, 1996). Interviews are particularly useful for getting the story behind a participant's experiences. The interviewer can pursue in-depth information concerning the research topic in order to follow-up on certain responses to questionnaires (McNamara, 1999).

Interviews are based on what the respondent says, and are therefore a far more personal form of research than questionnaires. In a personal interview, the interviewer works directly with the respondent and has the opportunity to ask follow up questions. Generally they elicit responses with greater ease, especially if what are sought are opinions or impressions. On the other hand the flexibility of interviews can bring dangers in that certain topics may be overlooked or may be addressed in an inconsistent order. Moreover, the interviewer may lead the respondent, shaping responses through the tone in which questions are asked or non-verbal gestures, and the responses may become as complex as to make analysis very difficult if not impossible. Given the time interviews take, it is difficult to interview large numbers of respondents.

There are three types of interviews: structured; semi-structured and unstructured (or in-depth) interviews. The structured interview is determined by the extent to which the questions are fixed prior to the interview; questions are pre-set, and closed in nature. The responses may also be pre-coded allowing for the collection of quantitative data.

Structured interviews can be replicated with multiple interviewees. The data produced tend to be more representative of the population than semi-structured or unstructured interviews and are generally easier to analyse. Usually the questions are asked in a specific order allowing the interviewer to maintain maximum control over the interview.

Semi-structured interviews: In a semi-structured interview, the interviewer will have some predetermined questions but can also ask some spontaneous questions. The predetermined questions tend to form a guide to the interview; they do not dictate the course of the interview and tend not

to have a set order. As some predetermined questions are used, quantitative data can still be collected and some coding can still be used. Consideration needs to be given to how data produced from spontaneous questions will be handled, especially in cases where spontaneous questions may be asked of some interviewees and not others.

In unstructured interviews, the interviewer does not have predetermined questions beyond the opening question but the interview will still revolve around a theme or topic and the researcher needs a clear idea of the depth of information that is needed so that useful information can be gathered. An unstructured interview format allows the interviewer to be responsive to the interviewee making the interview more conversational in style. The interviewer's control of the interview may therefore be limited. The data produced is completely qualitative which makes it difficult to replicate. Unstructured interviewing can be a time consuming way to generate data. There is also greater potential for interviewer bias using this type of interview.

In this research, unstructured interviews were used because no predetermined questions were asked (Curry et al., 2009). The reason for using this type of interview is to remain as open and adaptable as possible to the interviewee's qualities and priorities. The research interview was a recorded, open interview that was conducted only with the parents of children with TS. The interviewer presented one initial descriptive question aimed at eliciting as much information as possible: *'Describe what it is like to live with a child with TS at home'*. A single open question was used so that the researcher could provide the family with the opportunity to continue to develop the discussion as they desired, and to indicate what they felt was important, without any intervention or direction on the part of the researcher. The goal was to understand their personal experience as they felt it and the meaning that they attributed to this experience. Both the researcher and the family contributed to and were affected by this process of re-constructing reality (Woods & Marcks, 2005). This allowed better and deeper understanding of the impact that the child with TS has on the FQOL of the entire family.

4.6 Reliability, Validity and Generalisability in Qualitative Research

According to Mason (2006:21) '*... validity, reliability, and generalisability are different kinds of measures of the quality, rigour, and wider potential of research, which are achieved according to certain methodological and disciplinary conventions and principles.*' 'Reliability' and 'validity' are therefore terms used to address the quality of the data and appropriateness of the methods used in carrying a research project. The quality of the data and the appropriateness of the methods employed to collect them are particularly important in the social sciences because of the different, not necessarily scientific, philosophical and methodological approaches to the study of human activity.

4.6.1 Reliability

'Reliability' relates to the consistency of measurement, or the degree to which an instrument measures phenomena the same way each time it is used under the same condition with the same subjects. In short, it is the repeatability of the research measurement. If the method of collecting and analysing data is repeated, then the claims and the conclusions should be the same. Moreover, researchers should provide detailed information regarding the ways the evidence was gathered and analysed to enable another researcher to follow these procedures, and have confidence in them. Reliability addresses the accuracy of the research methods and techniques that produce the research data.

The use of multiple methods in a research in order to corroborate data sources increases the reliability. **Triangulation** tests the consistency of findings obtained through different instruments. Triangulation of several data sources and different methods increases the reliability of the data and of the interpretation of the data (Bryman, 2006) and confirms the validity of the processes (Yin, 1989).

In this research, triangulation of data from different sources (Denzin & Lincoln, 2000) increased chances to control, or at least assess, some of the threats to validity or deviations influencing the results. As mentioned before, both questionnaires and interviews were used in the present research, thus qualitative sources of evidence relating to the families of children with TS, produced information about the FQOL of these families that qualified the quantitative scores and statistics.

4.6.2 Validity

A test of 'validity' investigates whether research explains or measures what it intended to explain or measure. It therefore deals with the appropriateness of the method to produce responses to the research question. Here, the question to be asked is: *'how well matched is the logic of the method to the kinds of research questions you are asking and the kind of social explanation you are intending to develop'* (Mason, 2006:147). This is the test that justifies the appropriateness of method to the research question.

Validity relates to the validity of the interpretations of the data. All interpretations are subjective, so the issue here lies in tracing the ways to arrive at this particular interpretation. According to Mason (2006), a test of validity investigates whether the researchers are observing, identifying, or 'measuring' what they say they are. However, the relevance of these concepts to qualitative research has been a source of debate among qualitative researchers. Reliability estimates the consistency of measurement. In contrast, validity, involves the degree to which we are measuring what we are supposed to. In this sense, validity is more important than reliability in a qualitative research because if an instrument does not accurately measure what it is supposed to, there is no reason to use it even if it measures consistently. However, measurement validity is sometimes considered of little concern to qualitative researchers, since measurement is not an inherent issue in such researches.

Internal validity refers to the good match that exists between the researchers' observations and the theoretical ideas they develop. This quality reflects the strength of a qualitative research.

External validity refers to the degree to which a study can be replicated; it denotes the degree to which findings can be generalised across social settings. According to LeCompte and Goetz (1982), external validity is difficult to achieve in qualitative research, because of its tendency to use case studies and small samples, and because social settings are not replicable (ibid).

In the present study, reliability of the quantitative data is high, whereas the validity of the quantitative data is relatively low. Additionally, for the qualitative data reliability is low and validity is high.

4.6.3 Generalisability

Generalisability is defined as the '*degree to which the findings can be generalised from the study sample to the entire population*' (Polit & Hungler, 1991:645). According to Adelman et al. (1980), the knowledge generated by qualitative research is significant in its own right. In addition to concerns about generalisability, qualitative methodology is rebuked because studies are often difficult to replicate. The mission of qualitative research is to discover meaning and understanding, rather than to verify truth or predict outcomes. Therefore, in this inductive research, that aimed to develop an understanding of perceptions pertaining to FQOL of families who have a child with TS, reliability is low and validity is high. It should be noted that this is a small scale research. Therefore, despite the careful and rigorous manner in which this was undertaken, it is difficult to make well-founded claims regarding generalisability. Nevertheless, it is anticipated that the findings and conclusions of the research may be significant for the formation of future family policy and practice.

4.7 The Research Population and Sampling

Sampling is the procedure a researcher uses to gather people, places, or subjects to study. The sampling approach that is used in this research is a convenience sample. This means that the researcher selected a group of people for participation who were readily available, according to the specific characteristics that the researcher was interested in investigating. Such samples use individuals who are available rather than selecting from the entire population (Lunsford & Lunsford, 1995; Johnson & Turner, 2003). In this case, the researcher made no attempt, or only a limited attempt, to ensure that this sample was an accurate representation of a larger group or population. Convenience samples often have great difficulty in generalising the results to any population that has practical relevance, therefore, in order to interpret the findings from a convenience sample properly, it is necessary to characterise (usually in a qualitative sense) how the particular sample employed would differ from an ideal sample that was randomly selected.

The general research population included all families of children with TS in Israel. The specific research population that was approached to participate in the research included all those families in Israel at the time of the research that had a child with TS living in the family home, who were registered with the TS Organisation in Israel (TSOI). The participants were identified with the help of the TSOI. The TSOI staff mailed letters to the families requesting their agreement to participate in the research. All families who agreed to participate in the research were included as participants (25 Jewish families) and these participants completed the questionnaires.

Only ten families out of the 25 families with children with TS who responded to the questionnaires agreed to also participate in the interview. At this stage it is noted that the group of families who consented to participate in the research were homogenous in ethnic origin although the population from which they were drawn (all those families registered with the TSOI) is not so homogenous from this aspect. All families who agreed to participate in the research were included as participants (25 Jewish families) and they completed the questionnaires as a convenience sample. Israel is a multi-

cultural society, including various ethnic groups, such as Muslims, Christians, Druze, and Circassians. The reasons why only Jewish families responded to this study could be that members of other ethnic groups are insufficiently aware of the existence of TSOI and its significance, or reluctant to be exposed as the parents of a child with TS. It could also reflect the families' reluctance perhaps to engage with structures that they perceive to be insensitive, inflexible and personally exposing. However, the reasons for this were not investigated and this fact may constitute a reason for bias in the research results. Table 1 below presents a summary of the general attributes of the research samples.

Table 1: Summary of General Attributes of the Research Samples

Respondent	Variable	Values	Families of children with TS		Total	
			F	%	f	%
Father	Age group	30-40	4	19.0	7	21.2
		41-50	6	28.6	13	39.4
		50+	11	52.4	13	39.4
	Employed	Full time	19	90.5	30	90.9
		Part time	1	4.8	1	3.0
		Unemployed.	1	4.8	2	6.1
	Education	No. Matriculation	0	0.0	1	3.0
		Matriculation	8	38.1	11	33.3
		Higher Education	2	9.5	2	6.1
		B.A.	6	28.6	9	27.3
		M.A. +	5	23.8	10	30.3
Mother	Age group	30-40	5	20.0	8	21.6
		41-50	14	56.0	22	59.5
		50+	6	24.0	7	18.9
	Employed	Full time	13	52.0	24	64.9
		Part time	7	28.0	8	21.6
		Unemployed.	5	20.0	5	13.5
	Education	No. Matriculation	1	4.0	1	2.7
		Matriculation.	13	52.0	15	40.5
		Higher education	3	12.0	5	13.5
		B.A.	6	24.0	10	27.0
		M.A.+	2	8.0	6	16.2
Siblings	Gender	Male	8	50.0	16	48.5
		Female	8	50.0	17	51.5
	Age group	5-10	1	6.3	6	18.2
		11-15	6	6.3	9	18.2
		16-20	5	31.3	12	36.4
		21+	4	25.0	6	18.2
	Education	Elementary	5	31.3	12	36.4
		No. Matriculation	2	31.3	2	36.4
		Matriculation.	4	25.0	9	27.3
		Higher Education	1	6.3	2	6.1
		B.A.	3	18.8	7	21.2
		M.A.+	1	6.3	1	3.0

Respondent	Variable	Values	Families of children with TS		Total	
			F	%	f	%
Family	Income	Much higher	7	29.2	16	45.7
		Higher	5	20.8	6	17.1
		Equal	4	16.7	5	14.3
		Lower	5	20.8	5	14.3
		Much lower	3	12.5	3	8.6
	Members	1-3	7	28.0	7	18.9
		4-5	14	56.0	23	62.2
		6+	4	16.0	7	18.9
	Status	Married	22	88.0	34	91.9
		Single mother	3	12.0	3	8.1

Table 2 below provides details regarding the characteristics of the children with TS who participated in the research with their families.

Table 2: Background Data of the Children with TS

Variable	Values	Number	Percent	Net Percentage
Age Group	6-10	11	29.7	44.0
	16-20	11	29.7	44.0
	21+	3	8.1	12.0
	Total Valid	25	67.6	100.0
	Missing Values	12	32.4	
	Overall Total	37	100.0	
Gender	Boy	20	54.1	80.0
	Girl	5	13.5	20.0
	Total Valid	25	67.6	100.0
	Missing Values	12	32.4	
	Overall Total	37	100.0	
Disability Level	Mild	3	8.1	12.0
	Medium,	8	21.6	32.0
	Severe	14	37.8	56.0
	Total Valid	25	67.6	100.0
	Missing Values	12	32.4	
	Overall Total	37	100.0	
Secondary Disability	Yes	21	56.8	87.5
	No	3	8.1	12.5
	Total Valid	24	64.9	100.0
	Missing Values	13	35.1	
	Overall Total	37	100.0	

4.8 Ethical Considerations

The sample group included families who had a child with TS (parents and siblings). The families were chosen with the help of the TS Organisation in Israel (TSOI). The organisation obtained the families' written approval to participate in the research. I contacted the families through the organisation.

No other procedures were carried out with the subjects, apart from the questionnaires and interviews. The researcher assured the families that no harm would come to the respondents during the interviews and that they could withdraw at any time. Additionally, the researcher did not envisage any potential risks to the participants as a result of their response to the questionnaire and the interview. However, if the respondent felt uncomfortable or distressed, the interviews stopped. All participants had the right to withdraw or refuse their informed consent. If the siblings were under the age of 18, the parents signed their consent form. If they were older than 18, they signed their own consent form.

4.8.1 Anonymity

The issue of anonymity was discussed by the researcher with the informants when it came to their names being used in the final draft of the dissertation. Pseudonyms were used instead of their actual names. After the interview was completed, the researcher asked the participants if there was any material that they did not feel comfortable with the researcher using, and this section was marked to ensure that it was not transcribed at any point.

4.8.2 Confidentiality

This issue was also discussed with the participants before the interviews took place. Firstly, the participants received an explanation of the way in which the information would be maintained in strict confidentiality. This was performed by locking all tapes, transcripts, notes and any other material gathered during the interview in a lockable filing cabinet at the researcher's home. During transcription of the data, the pseudonyms were used and the key to their use was also locked in the filing cabinet at the researcher's

home. The researcher informed the informants that the information collected at the interviews and the questionnaire would be available to them upon their request and offered to show them the entire dissertation whenever they asked for it. The researcher also discussed with them what would happen to the data collected from them, after the project had finished. In addition, the researcher also discussed the possibility of keeping the data for future research (locked in a secure place or archived on a protected CD). If they had any objections to this then their data would be destroyed, including all tapes, notes and transcripts.

The researcher informed the participants that they could withdraw at any time, ask for data to be destroyed at any time and that they had the authority to read the dissertation at any time.

All the above explanations were recorded on an ethical consideration sheet for each informant and attached to their notes and transcripts.

The consideration of ethical issues, described above, conformed strictly to the Anglia Ruskin University research ethics code (Anglia Ruskin University, 2004-2005).

4.9 The Researcher's Role and Level of Involvement

The TS Organisation in Israel (TSOI), where families of children with TS are registered as members, sent letters to 450 registered families (all the families with a family member with TS in Israel), and attached a letter written by the researcher, asking them to participate in the research. Those who were interested in participating in the research were asked to contact the researcher by telephone or via the Internet.

Thirty families contacted the researcher and expressed their willingness to participate in the research. Five families were unsuitable for the selection criteria. In two of the families, the child with TS did not live at home, in one family the child was already 30 years old, and in one family, they were uncertain whether their child had TS, and another family a family member did not want to complete The questionnaire. The research population was distributed throughout the country, from Be'er Sheva in the South to Tiberius in the North.

The Questionnaire

The researcher requested permission to use the Beach Centre FQOL Survey questionnaire, from the Beach Centre in Kansas University, U.S.A. and received their written authorisation to use the questionnaire. The Beach Centre sent an original questionnaire via e-mail (see the request letter and the original questionnaire in Appendix 1).

Distribution of the Questionnaire

Families who agreed to participate in the research signed a consent form which included the researcher's promise to maintain confidentiality (the consent form appears in Appendix 4).

The researcher distributed the questionnaires personally, a task which required a great deal of time and hard work. This strategy was chosen because of the researcher's desire to oversee the process personally, verifying that the questionnaire was delivered to the families in a proper

manner, with a uniform explanation provided to everyone in order to ensure that each of the parents and siblings filled in the questionnaires themselves, and that there was no interference from other people.

Each family received an explanation regarding the research aim and instructions regarding the filling in of the questionnaire. If questions arose, the researcher explained the issue and asked the respondents to respond as best they could. The researcher avoided providing too many explanations to prevent any deviation that the explanation might engender.

The researcher contacted each family by telephone to arrange the meetings. She focused on different areas of the country separately. It was very difficult to set up meetings since some families wanted to meet in the morning hours when the children were at school. Others wanted to meet in the afternoon because they worked during the day.

Most of the families asked the researcher to visit when the children were not at home, which made it difficult to obtain the participation of siblings in the research. Therefore, in some families, the siblings did not fill in the questionnaire.

After filling in the questionnaire, the researcher asked the parents if they were willing to be interviewed. She set up another meeting on a different day with the families that agreed to be interviewed (ten families). The data from the questionnaires were directly inputted into an Excel file which was prepared for the purposes of the research data, directly after each questionnaire was completed in order to prevent a heavy workload at the end of the process.

The Interviews

The researcher conducted all the interviews. The interviews took place in the homes of the respondents two weeks after completing the questionnaire, and lasted 60-90 minutes each. The researcher explained the background of the study to the interviewees and explained the importance of the study. The researcher explained that the interviews would be recorded and would be

confidential, and received permission to do so. The interview was conducted with both parents at the same time. An open descriptive question was posed: *'Describe what it is like to live with a child with TS at home'*; and the parents were allowed to answer as they desired. The researcher did not interfere in the ensuing conversation. In some families, the mother gave most of the information, and in some families the answer was shared. For example: the mother said one thing, the father continued, and so forth.

The interviews were recorded on a digital tape recorder and transferred to a computer file. Then the content was transcribed verbatim, without any changes or editing.

4.10 Methods of Analysis

4.10.1 Introduction

The present research includes the investigation of indices relating to four areas of interest: Participants' Personal Characteristics, Quality of Life (QOL), services for the child with TS and services for the family. For each one of these groups of variables, data were gathered from the father and the mother. By calculating the average of the father's and mother's indices, a group of parental indices was created. Indices for the QOL and services for the family were also gathered from the siblings of the child with TS, and calculation of the average of the responses of the siblings yielded a 'siblings' index. At the end of the process, the average for all respondents (father, mother, first and second sibling), was calculated to create a 'family' index. The following is the analysis of the indices by groups of variables

Personal Variables

The present research investigated parameters from four groups of variables: Personal characteristics, QOL, services to the child with TS, and services to the family. Data concerning each one of these groups of variables were gathered from five different family members: the child with TS, the father, the mother, and siblings (one or two).

Personal Characteristics:

1. Age of respondent - this variable was measured in complete years was and was recorded for each of the respondents (the child, father, mother, and siblings).
2. Gender - The gender of the respondent (male/female). This variable exists for the respondents (the child, father, mother, and siblings).
3. Education level - the level of education of the respondent was noted on a scale of six levels for the parents and siblings (see question 4, Part Four of the questionnaire, Appendix 1) .This variable exists for the respondents (father, mother, and siblings).

4. Employment status - the respondent's employment status was noted on a scale of four levels, this variable exists for the parents (see question 3 of part Four of the research questionnaire).
5. Degree of disability: The child's degree of disability based on the mother's report. This variable originally contained three levels (mild, intermediate, severe). For the purposes of the present research, the two milder degrees were unified leaving two levels: mild/intermediate (1) and severe (2). (See question 8, Part Four of the questionnaire, Appendix 1).

General Details Variables

1. Age of respondent - the average ages of the partners in the family (parental, sibling). (See statement 2, Part Four of the Research Questionnaire, Appendix 1).
2. Income level - the assessment of average income of the mother and father, in relation to the average income in Israel. This assessment is rated on five levels, with 1 being much lower than the average income in Israel and 5 being much higher (see statement 10, Part Four of the Research Questionnaire Appendix 1).
3. Number of family members - derived from the mother's report of the number of individuals in the household (see statement 11, Part Four of the Research Questionnaire, Appendix 1).
4. Family status - derived from details in the mother's report regarding the family household status (see statement 3, Part Four of the Research Questionnaire, Appendix 1).

Quality of Life Variables

These variables are data which evaluate the family's level of satisfaction with actual FQOL as assessed by the various respondents, as detailed above. There are two types of basic indices for these variables, as measured in part 3 of the Research Questionnaire (Appendix 1) - the importance of the QOL domain and level of satisfaction. In the 'importance of the domain' scale, the assessment runs from 1 to 5, where a high rating on the index (5) testifies to a high degree of importance. In the 'satisfaction'

scale, the values run from 1 to 5, where a high rating (5) testifies to high satisfaction. Additionally, a calculation was made of the difference between each of the indices on the importance scale and the parallel index on the satisfaction scale.

The possible range of values for calculation of the difference runs from 4 to (-4), with a high rating (4) testifying to high satisfaction relative to the importance of the area. Table 3 below lists the indices included in this area:

Table 3: Itemisation of FQOL Parameters

Parameter	Items	Internal Reliability		Notes
		Importance	Satisfaction	
Family Interaction	1, 3, 7, 10, 11, 12, 18	0.75	0.86	
Parenting	2, 5, 8, 14, 19	0.70	0.75	
Emotional well-being	4, 9, 17, 21	0.73	0.69	
Physical well-being	6, 13, 15, 16, 20	0.85	0.82	
Support for child with TS	22, 23, 24, 25	0.69	0.74	Doesn't appear in siblings and family

Variables relating to Services for the Child with TS

These variables assess the degree of necessity of services for the child with TS and the degree to which they were received as assessed by the mother and father of the child. The indices for this area are calculated as the average of the items in Question 2 of part 1 of the Research Questionnaire, (Appendix 1) as assessed by the mother, father and parents (average of the mother's and father's scores). There are two basic types of indices for these variables as measured in the questionnaire - degree of necessity of the service, and degree of service actually received. On the 'degree of necessity' scale, the values run from 0 to 1, with a higher rating (1) testifying to a strong necessity for the service.

On the 'service actually received' scale, the values run from 0 to 2, with a higher rating (2) indicating a more complete service received. Additionally, the difference between the 'necessity' and 'service actually received' indices was calculated for each variable. The possible scale of values for this index runs from 1 to (-) 1, with a higher rating (1) testifying to a more complete service received relative to the necessity of the service. Table 4 below itemises the indices included in this category.

Table 4: Itemisation of 'Assistance for Child' Parameters

Parameter	Items	Internal Reliability		Notes
		Necessity	Service received	
Equipment	1, 9	0.74	0.71	N/A for siblings and family
Health	2, 3, 4, 5	0.82	0.79	N/A for siblings and family
Education	6, 10	0.72	0.74	N/A for siblings and family
Psychology	7, 8	0.71	0.70	N/A for siblings and family
Coordination	11, 12	0.72	0.76	N/A for siblings and family
Employment	13	--	--	N/A for siblings and family

Family Services Variables

These variables provide an assessment of the degree to which services were received for the family of the child with TS as assessed by all respondents (father, mother, parents, siblings, family), and they are calculated as the average of the items of part 2 of the Research Questionnaire (Appendix 1). In the indices for this area, there are two basic types of indices as measured in the questionnaire, degree of necessity of the service for the family and degree of service actually received. On the 'necessity' scale, the values run from 0 to 1, with a higher rating (1) testifying to a strong necessity for the service. On the 'service actually received' scale, the values run from 0 to 2, with a higher rating (2) indicating that a more complete service was received. The difference between the 'necessity' and 'service actually received' indices was calculated for each variable. The possible scale of values for this index runs from 1 to (-) 1, with a higher rating (1) testifying to more complete service received relative to

the necessity of the service. Table 5 below itemises the indices included in this area.

Table 5: Itemisation of 'Assistance for Family' Parameters

Parameter	Items	Internal Reliability	
		Necessity	Service received
Equipment	1, 4	0.66	0.69
Family	2,4	0.69	0.71
Money	3	--	--
Support/Counselling	6, 7, 8, 9	0.72	0.73
Information	1, 11, 12, 13	0.72	0.71

4.10.2 Data Analysis Procedure

Statistical processing

Statistical data analysis was deemed appropriate for the central research concept and the hypotheses stemming from it. The central analyses were comparative analyses between items and between family systems regarding QOL and provision of services. These analyses focus on the use of statistical tools which analyse differences between dependent variables (t-test for paired groups). In addition, Pearson correlation coefficients were calculated.

To start the data analysis, a number of descriptive analyses were conducted, including frequencies, means, and standard deviations, in order to understand the distribution of the research variables. At the end of the research between-group analyses were conducted in the form of t-tests for independent groups to examine the differences between the variables among families who have a child with TS (the degree of the child's disability varied).

Analysing interviews

In qualitative research the data analysis is almost all interpretive. It is not an accurate representation, as in positivistic traditions, but more a reflexive reactive interaction between the researcher and the *'de-contextualized data that are already interpretations of a social encounter'* (Cohen et al., 2000:282). The role of the researcher in analysing interviews is to maintain the holistic nature of the interview in relation to the units of meaning that are extracted from the interview to serve the purpose of the research. There are several stages in analysing interviews:

1. Generating natural units of meaning.
2. Classifying, categorising, and ordering these units of meaning.
3. Structuring the narratives to describe the interview contents.
4. Interpreting the interview data.

Content analysis is a standard methodology used in the social sciences to study the content of communication. The method of content analysis used enables the researcher to include large amounts of textual information and systematically identify its properties, e.g. identifying the frequencies of the most frequently used keyword by detecting the more important structures of its communication content (Holsti, 1969). Yet this textual information must be categorised according to a certain theoretical framework, which informs the data analysis, eventually producing a meaningful reading of the content under scrutiny. In this research, interviews were analysed using content analysis, relating to words and descriptions provided by the respondents, reflecting their feelings, thoughts, beliefs, desires and knowledge. Content analysis is a systematic research method for analysing textual information in a standardised way that allows evaluators to make inferences about that information (Weber, 1990, pp.9-12, and Krippendorff, 1980, pp. 21-27). *'A central idea in content analysis is that the many words of the text are classified into much fewer content categories'* (Weber, 1990, p. 12). Creswell (2003) maintains that the process of data analysis includes making sense of texts. It is a reflexive reactive interaction between the researcher and the de-contextualised data that are already interpretations of a social encounter (Cohen et al., 2003:282). As noted by Robson (1993:370), there

is' no clear and accepted set of conventions for analysis corresponding to those observed with quantitative data.'

The data analysis in this research was conducted in 7 stages (Miles & Huberman, 1994; Kassan & Krumer- Nevo, 2010).

1. Transcribing the interviews- (Appendix 5)
2. Coding- Dividing the data into meaningful segments and giving names to similar phenomena –(Appendix 5)
3. Mapping Stage - Creating a 'Category Tree': involving a review of the rationale for coding and an identification of the emerging patterns and themes. Attempting to attribute the categories to the FQOL domains.1. Family interaction 2. Parenting 3. Emotional well-being 4. Financial well-being 5. Support for the child and family (see page 137-150).
4. Incubation - reflecting on the data, and developing interpretations and meanings (see page 137-150).
5. Culling, condensing and reinterpreting the data so that they can be written up intelligibly.
6. Interpretation - extracting meanings of the data.
7. Conceptualising and writing - The Theoretical Stage- Through reflection I conceptualised the main themes from commonly used phrases transforming them into theoretical and conceptual issues and findings.

More specifically, the interviews were transcribed and then scanned in order to detect categories that could be connected to the five FQOL domains. Using colour highlighting techniques, the phrases and expressions pertaining to the different domains were marked on the text, and then categorised according to the different QOL domain columns. For example, the phrase: *'There were many problems and much tension'* was interpreted as relating to 'Family Interaction'. Interestingly, some expressions could be related to two different domains. For example, when a mother talked about the difficulties of a sibling of a child with TS, she said *'only after I spoke to him I understood what was wrong'*; this could be considered as either

'Family interaction' or 'Parenting'. The decision was made to assign this expression to 'Family interaction' because the researcher interpreted it as an interaction and as representing the ability to be open and talk about a problem more than a parent giving support to a child. The relevant quotations are presented in this chapter as they emerged in the respondents' words, but with pseudonyms for those expressing these phrases in order to preserve the privacy and anonymity of the interviewees. The categories emerged as a result of this process, and are presented as findings in Chapter 5: Research Findings.

4.11 Limitations of the Research

Several limitations need to be addressed with regard to this study. Those limitations include research related biases, researcher related biases, research tools related biases, and generalisability of the findings.

1. The research population includes only families that provided their consent to participate in the research. Therefore, the research cannot claim to have comprised a representative sample of the entire population of families having a child with TS.
2. The present research relates to the entire Israeli Jewish population who lived with their child with TS and who agreed to participate in the research. There was no limitation placed on place of residence, religion, ethnic group or socio-economic status. Although this decision provided the research with maximal reliability, it also caused logistic difficulties because of the large distances between the homes of the various families. There were families living in the Galilee in the north of the country, and families at the southern end of the country, in Beer Sheba.
3. The researcher preferred to conduct the research herself. Therefore, she often had to travel more than once to families that agreed to be interviewed. Each interview lasts between 60 to 90 minutes. This necessitated much time to gather the data. On the other hand, it provided depth and interest to the research.

4. In some cases, in families that have consented to participate in the research, one family member did not agree to complete the questionnaire, and therefore the entire family did not participate. This affected the quality of data gathering.
5. Some of the families, who agreed to participate in the research, changed their minds. I attempted to ask them to participate in the research via telephone (but in some cases this did not alter their decision).
6. The research related to the concept of the nuclear family (father, mother, sister, brother), and not just anyone living in the home. It would have been preferable to relate to everyone living in the home, in the research. Further research may find it valuable to examine additional circles affected by the existence of a child with TS in the family, such as: grandparents, and perhaps even uncles, and other relatives.

4.12 Summary

The methodology chapter provided a comprehensive discussion of the research approach that was adopted for this research, and the appropriateness of the methodology for the aims of the study was explained. Additionally, the research design and the research tools were presented, as well as the research population that provided the necessary data for achieving the aims of this study. Furthermore, issues related to validity, reliability, and generalisability were discussed, and the methods by which data were analysed were depicted. Finally, the ethical considerations that guided the research and the limitations of this study were presented. The following chapter will introduce the findings that emerged from the various research tools.

Chapter 5: FINDINGS

5.1 Preview

Chapter 5 presents the findings of the research, in the order of their emergence, in relation to the conceptual framework of this research, in order to strengthen the arguments towards their discussion in Chapter 6: Discussion. It presents the evidence as it emerged from the research instruments i. e., the questionnaire and the open interviews, and explains how the data were analysed to produce the findings of this research. The results of the analysis are presented in accordance with the research questions.

The main findings are then summarised through an account of the outcomes of the triangulation of data derived from the interviews and data derived from the questionnaires. In conclusion, this chapter prepares the grounding for annotations and interpretation of the findings that are discussed in Chapter 6: Discussion.

5.2 Findings emerging from the Questionnaire

5.2.1 Introduction

This section presents the findings as they emerged from the FQOL questionnaire. The aim of the questionnaire was to reveal how families who have a child with TS perceive their FQOL according to the five QOL domains (See Appendix 1 – Family Quality of Life Survey). The questionnaires were distributed to twenty-five families, and there was a 100% return rate. The data were analysed statistically in order to reveal the extent to which those families perceived that each of the five domains were important for their FQOL and to determine their order of significance.

Research Question 1

Which FQOL domains are perceived by family members to be affected by their child with TS?

In order to examine this question, a statistical analysis was conducted to evaluate the means and the standard deviations of different variables relating to individuals (father, mother) and variables relating to the different family systems (parents, siblings, family) living together with the child with TS. Statistical analyses were conducted to find the means and standard deviations of findings relating to the perceptions of importance of FQOL variables and level of satisfaction regarding these variables by individuals and families with a child with TS. Table 6 presents the analysis.

Table 6: Family Members' Perceptions of the importance of five QOL domains and the Level of their Satisfaction in these Domains

FQOL domains		Family		Siblings		Parents		Mother		Father	
		S.D	M	S.D	M	S.D	M	S.D	M	S.D	M
Family interaction	Importance	0.28	4.74	0.31	4.77	0.33	4.75	0.28	4.82	0.46	4.68
	Satisfaction	0.75	3.37	0.77	3.25	0.82	3.43	0.95	3.36	0.83	3.56
	Differences	0.81	-1.38	0.90	-1.53	0.85	-1.32	0.96	-1.46	0.92	-1.12
Parenting	Importance	0.37	4.46	0.40	4.60	0.39	4.45	0.41	4.50	0.49	4.37
	Satisfaction	0.64	3.43	0.71	3.42	0.71	3.47	0.82	3.40	0.74	3.61
	Differences	0.65	-1.04	0.73	-1.18	0.71	-0.98	0.82	-1.10	0.68	-0.76
Emotional Well being	Importance	0.35	4.47	0.34	4.46	0.41	4.48	0.44	4.46	0.48	4.46
	Satisfaction	0.60	3.36	0.73	3.54	0.62	3.32	0.68	3.24	0.72	3.46
	Differences	0.68	-1.11	0.58	-0.92	0.72	-1.16	0.79	-1.22	0.75	-1.00
Financial Well being	Importance	0.40	4.56	0.55	4.63	0.41	4.55	0.44	4.61	0.43	4.50
	Satisfaction	0.67	3.66	0.74	4.17	0.68	3.57	0.74	3.55	0.61	3.72
	Differences	0.76	-0.90	0.94	-0.45	0.75	-0.98	0.76	-1.06	0.66	-0.78
Support and services for the child	Importance	0.28	4.74	0.31	4.77	0.42	4.61	0.50	4.62	0.43	4.60
	Satisfaction	-	-	-	-	0.84	2.56	0.91	2.38	0.82	2.81
	Differences	-	-	-	-	1.02	-2.05	1.16	-2.24	0.97	-1.79

Table 6 shows that there was a difference in a negative direction between ratings for importance and degree of satisfaction for all FQOL variables and for all respondents (individuals and groups). This means that the reality of the FQOL fell short of expectations. It is also possible to see that for the *Family Interaction* and *Support for the Child* variables, there were relatively large differences, while in the *Physical Well Being* variable; there was a relatively small difference. Regarding standard deviations, there were relatively small deviations, indicating a very small variance between families. Therefore, the findings can be considered as reflecting a general tendency which is common to all the respondents and families, and not a tendency unique to specific families.

Thus, the finding shows that, even though there are large differences between degree of importance and degree of satisfaction for '*Family Interaction*' and '*Support for the Child*' all the families who had a child with TS perceived their FQOL domains in a similar manner.

Question 1A. Are there differences between the perceptions of mothers and those of fathers regarding the importance of the FQOL domains and their level of satisfaction in these domains?

In order to examine this question, paired sample t-tests were conducted to reveal differences in perception of the mothers' and fathers' regarding their FQOL and *Services for the Child and the Family*. The assumption behind this examination was that in light of the different functions of fathers and mothers in the family, it was possible that a significant difference would be found in their perceptions of the situation. Table 7 below presents the results of this analysis for each of the five QOL domains:

Table 7: Differences between Fathers' and Mothers' Perceptions of five QOL Domains

FQOL domains		Descriptive		Pearson		T test	
		S.D	M	R	Sig.	t	Sig.
Family interaction	Father	0.92	-1.12	0.51	0.02	1.61	0.12
	Mother	0.96	-1.44				
Parenting	Father	0.68	-0.76	0.38	0.09	1.51	0.15
	Mother	0.76	-1.03				
Emotional well being	Father	0.75	-1.00	0.45	0.04	0.84	0.41
	Mother	0.74	-1.14				
Financial well being	Father	0.66	-0.78	0.73	0.00	1.66	0.11
	Mother	0.63	-0.95				
Support for the child	Father	0.97	-1.79	0.57	0.01	2.13	0.05
	Mother	1.12	-2.24				

Table 7 indicates that for all the domains, mothers evaluated the difference between importance and satisfaction with FQOL as more significant than fathers did. Although none of the differences in the perceptions of the

variables between mothers and fathers were significant (although some are very close to significant, in particular for the domain *Support for the Child* – in this case relating to the child with TS), because of the consistency of this finding, it was possible to see a significant trend. An additional finding that is seen in the data is the level of the positive and significant correlation between the mothers' evaluations and the fathers' evaluations. This indicated that the assessment of FQOL was not personal but rather familial.

Thus, this finding suggests that the perception of FQOL was formulated on the family level, even though mothers perceive the domains more strongly.

Question 1B. What are the perceptions of father, mother and the whole family, regarding the services they perceive as necessary for the child(ren) and the services which are actually received for the child(ren) with TS?

In order to examine this question, a statistical analysis was made to evaluate the means and the standard deviations of findings for different variables relating to individuals (father, mother) and those relating to the whole family system living together with a child with TS. Table 8 below presents the analysis.

Table 8: The Level of Services needed and provided for the Child with TS as Perceived by Fathers, Mothers and Family

Services for the child		Family		Mother		Father	
		S.D	M	S.D	M	S.D	M
School Equipment	Necessity	0.21	0.13	0.22	0.12	0.22	0.12
	Received	0.28	0.08	0.28	0.08	0.30	0.10
	Differences	0.24	-0.05	0.25	-0.04	0.25	-0.02
Health Services	Necessity	0.28	0.38	0.28	0.35	0.28	0.40
	Received	0.19	0.22	0.24	0.23	0.22	0.19
	Differences	0.32	-0.16	0.31	-0.12	0.36	-0.21
Special Education	Necessity	0.27	0.24	0.29	0.22	0.30	0.29
	Received	0.26	0.13	0.27	0.14	0.27	0.12
	Differences	0.32	-0.11	0.31	-0.08	0.33	-0.17
Psychological Services	Necessity	0.42	0.67	0.43	0.68	0.46	0.67
	Received	0.52	0.40	0.65	0.48	0.45	0.36
	Differences	0.53	-0.27	0.61	-0.20	0.51	-0.31

Table 8 shows that there was a difference in a negative direction between necessity and services received for all the variables of service provision to the child with TS, and for all respondents (individuals and groups), although the difference is generally small. In general it may be said that the service provision to the child with TS falls short of expectations, although there is almost a balance between expectations and service provision in practice. This was seen particularly in the 'school equipment' index, where an almost complete balance was visible. A more severe difference was seen with regard to the *Psychological Services* variable, where the difference was greater than for the other variables. Between families, the standard deviations were relatively small, indicating low variance between families, and indicating that the findings reflected a general tendency and not a tendency unique to individual families.

Thus, this finding shows that, the families perceived a large gap between the need for *Psychological Services* for the child and the actual provision of services suggesting that families who have a child with TS feel that they do not receive the psychological services for the child that they need.

Question 1C. What are the perceptions of the father, mother and the whole family, regarding the services they perceive as necessary for the family and the services which are actually received for the family?

In order to examine this question, a statistical analysis was made to evaluate the means and the standard deviations of findings for different variables relating to perceptions of individuals (father, mother) and those relating to perceptions of the entire family living together with the child with TS. Table 9 below presents this analysis.

Table 9: Analysis of Perceptions of Level of Services needed and received by the Family of a Child with TS as Perceived by Fathers, Mothers and Family

Services for the family		Family		Mother		Father	
		S.D	M	S.D	M	S.D	M
Equipment	Necessity	0.33	0.48	0.36	0.44	0.35	0.55
	Received	0.30	0.20	0.31	0.16	0.37	0.26
	Differences	0.33	-0.28	0.29	-0.28	0.41	-0.29
Extended Family	Necessity	0.36	0.49	0.37	0.52	0.37	0.48
	Received	0.49	0.19	0.48	0.22	0.60	0.19
	Differences	0.47	-0.30	0.46	-0.30	0.54	-0.29
Money	Necessity	0.43	0.54	0.49	0.64	0.51	0.43
	Received	0.10	0.02	0.20	0.04	0.00	0.00
	Differences	0.44	-0.52	0.50	-0.60	0.51	-0.43
Support & Counselling Services	Necessity	0.27	0.72	0.27	0.74	0.28	0.73
	Received	0.35	0.25	0.37	0.26	0.39	0.27
	Differences	0.41	-0.48	0.43	-0.48	0.47	-0.45
Information	Necessity	0.28	0.76	0.28	0.79	0.33	0.71
	Received	0.27	0.15	0.32	0.18	0.27	0.14
	Differences	0.33	-0.61	0.32	-0.61	0.35	-0.57

Table 9 above clearly shows that there was a difference in a negative direction between necessity and received services for all FQOL variables and for all respondents (individuals and groups), as can be seen in Table 9. A significant difference was shown for the *Information* and *Support and Counselling Services* and *Extended Family* variables, indicating a severe perception of the situation, while a smaller difference was exhibited for the

Equipment and *Money* variables in which the level of service falls short of expectations, but there is almost a balance between expectations and provision of services in practice. There are relatively small standard deviations, indicating a very slight variance between families. It therefore seems that the findings reflect a general tendency, common to all the respondents and families, and not a tendency unique to particular families.

Thus, this finding suggests that, although 'information' and 'support and counselling services' were perceived as the most significant services needed, they were not perceived as being received to the extent needed, i.e. families who have a child with TS perceive that they do not receive the services for the family that they need.

Question 1 D. What are the differences between the perceptions of fathers and perceptions of mothers regarding the services they perceive as necessary for the child(ren) with TS and services for the family and the services which are actually received?

The difference between perceptions of fathers and mothers are presented in Table 10 and in Table 11.

Table 10: Differences between Fathers' and Mothers' Perceptions of the services necessary and received for the child with TS

Services for the child		Descriptive		Pearson		t-test	
		M	S.D	R	Sig.	t	Sig.
School Equipment	Father	-0.02	0.25	0.67	0.00	-0.57	0.58
	Mother	0.00	0.22				
Health Services	Father	-0.21	0.36	0.62	0.00	-1.50	0.15
	Mother	-0.12	0.27				
Special Education	Father	-0.17	0.33	0.84	0.00	-1.83	0.08
	Mother	-0.10	0.30				
Psychology Services	Father	-0.31	0.51	0.67	0.00	-1.67	0.11
	Mother	-0.14	0.59				

Table 11: Differences between Fathers' and Mothers' Perceptions of the services necessary and received for the Family

Services for the Family		Descriptive		Pearson		t-test	
		M	S.D	R	Sig.	t	Sig.
Equipment	Father	-0.29	0.41	0.75	0.00	0.00	1.00
	Mother	-0.29	0.25				
Family	Father	-0.29	0.54	0.81	0.00	0.00	1.00
	Mother	-0.29	0.46				
Money	Father	-0.43	0.51	0.48	0.03	1.71	0.10
	Mother	-0.62	0.50				
Support/ Counselling	Father	-0.45	0.47	0.68	0.00	0.15	0.88
	Mother	-0.46	0.44				
Information	Father	-0.57	0.35	0.67	0.00	0.20	0.84
	Mother	-0.58	0.29				

According to Tables 10 and 11 above, mothers and fathers gave identical evaluations for most of the variables. Fathers evaluated the situation regarding some of the variables as more severe than the mothers, while for others, the mothers evaluated the situation as more severe than the fathers. This may indicate that for all the service variables (for the child and the family) as a whole there was no significant difference between mothers and fathers.

Thus, this finding seems to reinforce the evidence from the previous table (Table 9) so that the findings suggest that the couples have a coordinated assessment of the situation, both with regard to services (for the child and the family) and with regard to their assessment of their FQOL. In other words, it seems that mothers and fathers share the same view regarding the assessment of 'services for the child and family' and FQOL.

Question 1E. What are the differences between the perceptions of their FQOL by parents of the child with TS and those of the child's siblings?

In order to answer this question, paired sample t-tests were conducted to ascertain the differences in perception of FQOL, between parents and siblings of the child with TS. The assumption underlying this examination is that in light of the different functions of parents and siblings regarding the child with TS, it was possible that a significant difference might be found in their perceptions of the situation. Table 12 below presents the results of this analysis in four FQOL domains.

Table 12: Differences in Perceptions of FQOL by Parents and Siblings

		Descriptive		Pearson		T test	
		M	S.D	R	Sig.	t	Sig.
Family Interaction	Parents	-1.24	0.85	0.77	0.00	1.75	0.11
	Siblings	-1.53	0.90				
Parenting	Parents	-0.96	0.70	0.36	0.22	0.97	0.35
	Siblings	-1.18	0.73				
Emotional Well-Being	Parents	-1.14	0.64	0.62	0.02	-1.48	0.16
	Siblings	-0.92	0.58				
Financial Well-Being	Parents	-0.94	0.64	0.41	0.16	-1.97	0.07
	Siblings	-0.45	0.94				

The findings of Table 12 above are in line with the findings of the analysis of the previous differences. Here too it was found that there was no significant difference between parents and siblings for any of the variables. However, for two variables (*Family Interaction* and *Parenting*) it was found that the siblings evaluated the situation as more severe, while for the other two variables (*Emotional Wellbeing* and *Physical Wellbeing*) the parents evaluated the situation as less severe than did the siblings.

Here too, as in previous analyses (between mothers and fathers), a strong, positive, significant correlation was found between perceptions of parents and siblings, reinforcing the understanding that the assessment of the situation was a family assessment expressed similarly by the father, mother and siblings, and not, as might be assumed, a personal assessment resulting from the different functions or status of individuals within the family.

Thus, this finding shows that, although the siblings had more severe perceptions of the importance and reality of the QOL domains of *Family Interaction* and *Parenting* the general perception of their FQOL seems to be common to all family members and formed holistically.

Question 1F: What correlations can be found between the fathers' perceptions of FQOL Domains and their perceptions of Services for the Child and Services for the Family?

In order to examine this question, Pearson's correlation coefficient analyses were calculated to reveal differences between fathers' perceptions of FQOL and fathers' perceptions of quality of services (for the child and family). It was assumed the fact that the family had a child with TS, had implications for the various variables, and therefore, it was reasoned that positive correlations would be found between the fathers' perceptions of FQOL and service variables. Table 13 below presents the results of the analysis.

Table 13: Correlations between the Fathers' Perceptions regarding Services for the Child and for the Family and Fathers' Perceptions regarding FQOL domains

Differences between the services			Differences in FQOL domains				
			Family interaction	Parenting	Emotional Well being	Financial Well being	Services For the child
Services For the Child	Equipment	Value	0.11	0.06	0.00	0.28	0.02
		Sig.	0.65	0.78	1.00	0.23	0.92
	Health	Value	0.55	0.63	0.54	0.37	0.34
		Sig.	0.01	0.00	0.01	0.10	0.13
	Education	Value	0.62	0.66	0.66	0.47	0.57
		Sig.	0.00	0.00	0.00	0.03	0.01
	Psychology	Value	0.63	0.57	0.60	0.30	0.29
		Sig.	0.00	0.01	0.00	0.19	0.20
Services For the family	Equipment	Value	0.26	0.46	0.43	0.50	0.48
		Sig.	0.25	0.04	0.05	0.02	0.03
	Family	Value	0.40	0.58	0.37	0.32	0.39
		Sig.	0.07	0.01	0.10	0.15	0.08
	Money	Value	0.16	0.34	0.23	0.26	0.04
		Sig.	0.48	0.13	0.32	0.25	0.85
	Support/Counselling	Value	0.67	0.63	0.57	0.40	0.50
		Sig.	0.00	0.00	0.01	0.08	0.02
	Information	Value	0.46	0.56	0.43	0.43	0.59
		Sig.	0.03	0.01	0.05	0.05	0.00

The findings presented in Table 13 above show that the correlations between fathers' perceptions of FQOL and fathers' perceptions of services for the child with TS and for the family were positive, as expected, but their strength and level of significance were inconsistent. It is clear that there was no correlation at all between perceptions regarding both *Family Services* and *Child Services* domains and perceptions relating to the variables of *Equipment* (services for the child) and *Money* (services for the family).

On the other hand, perceptions relating to a number of variables in the services domain were found to be significantly correlated to FQOL variables, such as *Education* (child services) and *Information* (family services). This means that the perception of FQOL was influenced by the perceptions of variables in the services domain: *Education* (child services) and *Information* (family services). Further, findings for a number of variables in the service domain were found to be correlated with findings for most of the FQOL variables: *Health and Psychology* (child services), *Support and Counselling*, and equipment (family services).

Thus, in light of these findings, it seems that in general, there is a correlation between the fathers' perceptions of some FQOL variables and their perceptions of services variables, but there were some variables for which no such correlation was found.

Question 1G. What correlations can be found between the mothers' perceptions of FQOL Domains and their perceptions of Services for the Child and Family

In order to examine this question, Pearson's correlation coefficient analyses were calculated to reveal differences between mothers' perceptions of FQOL and mothers' perceptions of family quality of services (for the child and family). It was assumed that the fact that the family lived with a child with TS had implications for the various variables, and therefore, it was reasoned that positive correlations would be found between perceptions of FQOL and perceptions of service variables. Table 14 below presents the results of the analysis.

Table 14: Correlations between Mothers' Perceptions of Services for the Child and for the Family and their perceptions of FQOL Domains

Differences between perceptions of the services			Differences in perceptions of FQOL domains				
			Family interaction	Parenting	Emotional Well being	Financial Well being	Services For the child
Services For the child	Equipment	Value	-0.03	0.23	0.14	0.61	0.07
		Sig.	0.89	0.27	0.50	0.00	0.73
	Health	Value	0.32	0.46	0.39	0.42	0.47
		Sig.	0.12	0.02	0.05	0.03	0.02
	Education	Value	0.50	0.49	0.39	0.33	0.45
		Sig.	0.01	0.01	0.05	0.11	0.03
	Psychology	Value	0.20	0.40	0.41	0.39	0.36
		Sig.	0.33	0.05	0.04	0.06	0.08
Services For the family	Equipment	Value	0.09	0.28	0.42	0.53	0.51
		Sig.	0.68	0.17	0.03	0.01	0.01
	Family	Value	0.25	0.45	0.55	0.62	0.66
		Sig.	0.23	0.02	0.00	0.00	0.00
	Money	Value	0.21	0.56	0.34	0.50	0.37
		Sig.	0.31	0.00	0.10	0.01	0.07
	Support/ Counselling	Value	0.35	0.40	0.66	0.41	0.64
		Sig.	0.09	0.05	0.00	0.04	0.00
	Information	Value	0.40	0.48	0.61	0.67	0.54
		Sig.	0.05	0.02	0.00	0.00	0.01

The findings featured in Table 14 above resemble the findings featured in Table 8, which show the correlations among the fathers. Here too, the correlations were positive, as expected, but were of mixed strength and significance. Perceptions of one variable in the services domain: *Information* (family services).were found to be significantly correlated to FQOL variables.

Thus, In light of these findings in general, there seems to be a correlation between the mothers' perceptions of different FQOL variables, but this is not significant, and is true only for most of these variables. It is also concluded that there are similarities in the correlations between the two different sets of variables found for fathers and for mothers.

Thus too, in light of these findings, it appears that in general, there is a correlation between some perceptions relating to FQOL variables and perceptions relating to services variables by mothers and fathers, but that no such correlations were found for some other variables.

Question 1H. What correlations can be found between the perceptions of both parents regarding the FQOL Domains and their perceptions of Services for the Child and Family?

Here too, in order to answer this question, Pearson correlation coefficients were calculated to reveal differences between perceptions of FQOL and perceptions of family quality of services (for the child and family) among the parents. The assumption behind this examination is that it was assumed that living with a child with TS had implications for the various variables, and therefore, it was reasoned that positive correlations would be found between perceptions of FQOL and perceptions of the different service variables. Table 15 below presents the results of these analyses.

Table 15: Correlations between the Perceptions of FQOL domains and Perceptions of Services for the Child and for the Family according to both parents

Differences between the services			Differences in FQOL domains				
			Family interaction	Parenting	Emotional Well being	Financial Well being	Services For the child
Services For the child	Equipment	Value	0.06	0.31	0.19	0.58	0.18
		Sig.	0.76	0.13	0.35	0.00	0.39
	Health	Value	0.57	0.71	0.56	0.46	0.50
		Sig.	0.00	0.00	0.00	0.02	0.01
	Education	Value	0.57	0.55	0.48	0.40	0.49
		Sig.	0.00	0.00	0.01	0.05	0.01
	Psychology	Value	0.48	0.61	0.54	0.45	0.38
		Sig.	0.02	0.00	0.01	0.02	0.06
Services For the family	Equipment	Value	0.20	0.39	0.45	0.53	0.57
		Sig.	0.33	0.05	0.02	0.01	0.00
	Family	Value	0.39	0.60	0.53	0.55	0.57
		Sig.	0.06	0.00	0.01	0.00	0.00
	Money	Value	0.22	0.59	0.36	0.49	0.30
		Sig.	0.29	0.00	0.08	0.01	0.15
	Support/ Counselling	Value	0.59	0.60	0.70	0.45	0.59
		Sig.	0.00	0.00	0.00	0.03	0.00
	Information	Value	0.56	0.61	0.67	0.66	0.72
		Sig.	0.00	0.00	0.00	0.00	0.00

The findings presented in Table 15 above, reinforce the findings in the previous tables which dealt with the correlations between fathers' perceptions of FQOL and their perceptions of services (Table 13) and the correlations between mothers' perceptions of FQOL and their perceptions of the different services (Table 14). Here too, it was noticed that the correlations were positive and quite strong. However, there were a number of variables for which the correlation was overwhelmingly significant, and others for which the correlation was not significant.

A significant correlation was found between the perceptions of *Services for the Child* and for the following QOL variables: *Health*, *Education*, and *Psychology*. Only for the *Equipment* variable, the correlation is not significant. A significant correlation was found between perceptions of the *Family Services* and the FQOL variables: *Equipment*, *Family*, *Support and Counselling*, and *Information*. Only for the *Money* variable the correlation is not significant. In summary, the general expectation that there would be a positive correlation between perceptions of FQOL variables and services variables was supported by the findings of the analyses, but not for all of the variables.

Thus, in light of these findings, it appears that in general, there is a correlation between perceptions of the services for the child and their perceptions regarding some FQOL variables, by both parents, although some variables were found not to be correlated. Perceptions of services for the family were found to be correlated with perceptions of FQOL.

Research Question 2:

How do different families raising a child with TS perceive their FQOL?

Question 2A: Do families of children with TS who perceive their child as having a severe form of TS have a different perception of their FQOL than families who perceive their child as having a mild form of TS?

It is noted that the families' perception of the severity of the child's TS was measured through responses to Part four: Questions 8 and 9 in the Research Questionnaire (Appendix 1). Question 8 measured parents' perceptions of the severity of their child's TS, and Question 9 measured the number of additional difficulties that the child had, such as OCD, ADHD, LD, etc. In order to answer this question, independent t-tests were conducted between families in which the child is perceived as having a severe case of TS and families in which the child is perceived as having a mild case of TS.

The expectation was that significant differences will be found between these two categories of families, because when a child has a more severe case of TS this makes the family life much more difficult, whereas when the child has a milder case of TS, the impacts on family life is less severe. The results of these analyses are found in Table 16 below.

Table 16: Differences in Perceptions of FQOL domains by Families according to Perceived Severity of their Child's TS

Respondent	FQOL domains	Perceived Level of Severity of the child's TS				t-test	
		Mild		Severe			
		M	S.D	M	S.D	t	Sig.
Father	Family interaction	-0.56	0.42	-1.62	0.96	3.35	0.00
	Parenting	-0.62	0.62	-0.89	0.73	2.04	0.06
	Emotional Wellbeing	-0.68	0.41	-1.30	0.88	2.10	0.05
	Financial Wellbeing	-0.86	0.74	-0.71	0.61	-0.51	0.62
	Support for the child	-1.45	0.65	-2.09	1.13	1.61	0.13
Mother	Family interaction	-0.74	0.67	-2.02	0.77	4.45	0.00
	Parenting	-0.64	0.73	-1.46	0.72	2.81	0.01
	Emotional Wellbeing	-0.73	0.59	-1.61	0.72	3.37	0.00
	Financial Wellbeing	-1.00	0.77	-1.10	0.78	0.32	0.75
	Support for the child	-1.61	1.08	-2.73	1.01	2.65	0.02
Parents	Family interaction	-0.63	0.40	-1.86	0.69	5.58	0.00
	Parenting	-0.61	0.55	-1.28	0.70	2.69	0.01
	Emotional Wellbeing	-0.68	0.36	-1.54	0.71	3.90	0.00
	Financial Wellbeing	-0.90	0.72	-1.05	0.80	0.49	0.63
	Support for the child	-1.47	0.82	-2.51	0.94	2.96	0.01
Siblings	Family interaction	-1.14	1.01	-1.98	0.54	1.90	0.09
	Parenting	-0.85	0.75	-1.57	0.53	2.03	0.07
	Emotional Wellbeing	-0.79	0.70	-1.08	0.41	0.95	0.37
	Financial Wellbeing	-0.18	1.09	-0.77	0.66	1.20	0.26
Family	Family interaction	-0.73	0.53	-1.88	0.61	5.06	0.00
	Parenting	-0.67	0.46	-1.32	0.65	2.95	0.01
	Emotional Wellbeing	-0.69	0.39	-1.45	0.68	3.53	0.00
	Financial Wellbeing	-0.72	0.69	-1.03	0.81	1.02	0.32

The findings presented in Table 16 above, show that assessment of FQOL is lower among families in which the child has a more severe case of TS, compared to families in which the child has a milder case of TS for all variables and among all types of respondents (with the exception of fathers' perceptions of the 'financial wellbeing' variable) This finding is in line with the basic expectations for this analysis. For most of the variables, these differences are significant, but not for all types of respondents.

Thus, these findings seem to indicate that the perceived severity of the syndrome may have an impact upon the FQOL of families having a child with TS.

Question 2B: Do families of children with what is perceived as a severe case of TS have different perceptions of the services variables for the family than families of children with what is perceived as a mild case of TS?

In order to examine this question, independent t-tests were conducted between families in which the child has a severe case of TS and families in which the child has a mild case of TS. The expectation was that significant differences would be found between the two categories of families, because when a child who has a more severe case of TS this phenomenon makes family life much more difficult, whereas when the child has a milder case of TS, this phenomenon impacts family life to a lesser extent. The results of the analysis are found in Table 17 below.

Table 17: Differences between Families in Perception of Services for the Family by Family Members according to perceived degree of Severity of the Child's TS

Respondent	Services for the family	Descriptive				T test	
		Mild and Medium		Severe		t	Sig.
		M	S.D	M	S.D		
Father	Equipment	-0.35	0.41	-0.23	0.41	-0.68	0.50
	Family	-0.35	0.34	-0.23	0.68	-0.53	0.61
	Money	-0.40	0.52	-0.45	0.52	0.24	0.81
	Support/ Counselling	-0.28	0.51	-0.61	0.39	1.20	0.24
	Information	-0.48	0.43	-0.66	0.26	1.17	0.26
Mother	Equipment	-0.32	0.25	-0.25	0.33	-0.59	0.56
	Family	-0.27	0.41	-0.32	0.50	0.27	0.79
	Money	-0.45	0.52	-0.71	0.47	1.29	0.21
	Support/ Counselling	-0.27	0.48	-0.64	0.31	2.23	0.04
	Information	-0.45	0.31	-0.73	0.29	2.29	0.03
Parents	Equipment	-0.32	0.32	-0.25	0.35	-0.51	0.62
	Family	-0.30	0.37	-0.30	0.55	0.04	0.97
	Money	-0.41	0.44	-0.61	0.45	1.11	0.28
	Support/ Counselling	-0.26	0.43	-0.64	0.31	2.47	0.02
	Information	-0.44	0.34	-0.73	0.26	2.32	0.03

Table 17 above shows mixed results for the analyses which are not necessarily connected to the perceived severity of the child's case of TS. For some of the variables, there is in fact a difference in perceptions between the two categories of family in the expected direction, i.e., the families of children with a more severe case perceive the state of services received as lower than families with a child who had a mild to intermediate case of TS. This difference is consistent and significant for the *Support and Counselling* and *Information* variables.

On the other hand, for other variables and in particular for the 'equipment' index, there is a difference in the opposite direction to that which was expected, such that families with a less severe case of TS assess the level of services received as lower than do families with a more severe case.

Thus, this finding seems to corroborate the previous one, that the severity of the syndrome may have an impact upon the perceptions of FQOL of families who have a child with TS in the 'services for the family' domain, *Support and Counselling* and *Information* variables.

Question 2C: Is there a difference between families of children with what is perceived as a severe case of TS and families of children with what is perceived as mild case of TS in the indices of difference of services for the child?

In order to examine this question, independent t-tests were conducted between families in which the child has a severe case of TS and families in which the child has a mild case of TS. The expectation is that significant differences will be found between the families, as when a child has a more severe form of TS then the accompanying phenomenon will make family life much more difficult, whereas when the child has a mild case of TS, the phenomena will impact family life to a lesser extent. The results of the analysis are found in Table 18 below.

Table 18: Differences between Families in Perception of Services for the Child by Family Members according to perceived degree of Severity of the Child's TS

Respondent	Services for the child with TS	Descriptive				T test	
		Mild and Medium		Severe		t	Sig.
		M	S.D	M	S.D		
Father	School Equipment	-0.05	0.28	0.00	0.22	-0.45	0.66
	Health services	-0.08	0.35	-0.34	0.34	1.75	0.10
	Special education	-0.10	0.32	-0.23	0.34	0.88	0.39
	Psychology services	-0.15	0.53	-0.45	0.47	1.39	0.18
Mother	School Equipment	0.00	0.22	-0.07	0.27	1.43	0.17
	Health services	-0.02	0.13	-0.20	0.38	1.87	0.07
	Special education	0.05	0.15	-0.18	0.37	2.05	0.06
	Psychology services	0.00	0.59	-0.36	0.60	1.49	0.15
Parents	School Equipment	-0.02	0.24	-0.07	0.25	0.50	0.62
	Health services	-0.05	0.22	-0.25	0.36	1.24	0.23
	Special education	-0.02	0.21	-0.18	0.37	1.32	0.20
	Psychology services	-0.09	0.50	-0.41	0.53	1.54	0.14

The findings appearing in Table 18 above, indicate that almost consistently (with the exception of the *Equipment* variable among fathers) families of children perceived to have a more severe level of TS assessed the level of 'services for the child' that they received as lower in comparison to families of children perceived to have an intermediate to mild case of TS. This finding is in line with the main assumption underlying this examination. However, in none of the variables was this difference significant, so that it seems that the tendency for the difference exists, but is not sufficiently significant to allow clear conclusions.

5.3 Findings Emerging from the Open Interviews

5.3.1 Content Analysis

This part discusses the issues emerging from the interviews with parents of children with TS, and serves as the basis for the annotations and interpretation of the findings discussed in the next chapter, Chapter 6: Discussion.

In accordance with the research programme, data were also gathered from interviews of parents who have children with TS. Ten interviews were conducted and recorded in the families' homes; no more than two weeks after the questionnaires were conducted. The aim of these interviews was to capture the perceptions and feelings of those families regarding their experiences in raising a child with TS, in order to understand their FQOL more deeply through an examination of the different QOL domains (Turnbull et al., 2004), and how they coped with raising a child having TS on a daily basis. This chapter presents the findings that emerged from the content analysis of the interviews with those families. Analysis of the interviews focused on the five domains of FQOL as they appear in the research: family interaction, parenting, physical wellbeing, emotional wellbeing, and services for the child and the family.

Additionally, it is important to note that the analyses were performed immediately after the interviews were conducted, in order to retain as much information as possible. The researcher's personal impressions and notes were added to the interviews' analysis in order to enrich the data. Table 19 presents the results of the content analysis that was performed on the data emerging from the interviews, including sample evidence in the form of quotations.

Table 19: Findings emerging from the Interviews

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
FAMILY INTERACTION	<i>As if the family almost disintegrated. There were a lot of problems. In the house my husband and I are very close to one another, we have a very good relationship, but we were so tense and the tension was so hard on me. All the time, almost the entire time I was with him [the child with TS], like most of the time, I slept with him on the couch, because he would try to bite himself all the time and wound himself with his teeth, and I had to do reflexology with him, and it calmed him down and I would lie with him on the couch, and I would caress him until he would fall asleep. These were the nights that we slept on the couch. And my husband couldn't help.</i>	When the entire burden falls on the mother it causes a lot of tension.
	<i>The stress of our lives tore my husband and me apart, not an uncommon story in the life of a child with TS. We could not understand and we needed guidance to go through it all. We couldn't function as a family.</i>	When a parent cannot function a lot of stress is engendered.
	<i>Twice a week I go to exercise class and yoga and my husband puts the children to bed. It also helps. I can be by myself and it is very important to me, and I relax from the stresses of the day.</i>	Cooperation between parents helps alleviate pressure.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<p><i>At first my husband and I had difficult times. We did not know what to do, and this created tension between us. But now I feel that it has brought us closer to one another. My husband also has TS. We only found it out after we understood that our son has TS.</i></p> <p><i>Now we speak about it between us a lot and it helps us and makes us closer.</i></p>	The ability to talk and share with one another decreases stress.
	<p><i>I wanted to talk to my husband but I didn't know how to do so, I wish I could get some help.'</i></p>	Open discussions could help the parents deal in a better way with the situation.
	<p><i>'For a very long time my son did not invite any friends home. At first I did not understand that his behaviour was because of a feeling of shame and that he was afraid his brother would act strangely or even aggressively towards his friends'. 'When I decided to ask him about it, he told me that he was embarrassed by his brother's behaviour'.</i></p> <p><i>'My son did not invite his friends home because he did not want his friends to see his sister, because he thought he would get teased'</i></p> <p><i>'I thought that my brother was going to die because at first you would go off and discuss my brother's situation with my father in whispers.</i></p>	<p>Information about the syndrome can help siblings with their social and emotional life.</p> <p>Siblings needed information in order to maintain regular social life.</p>

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>No one told me anything, which caused me to think that something horrible was about to happen'.</i>	
	<i>When we discovered that our child likes to be out in nature, we began to plan hikes every week. The little children had a good time and he [the child with TS] was calm and happy. This brought the family closer together and allowed us quality time with the children.</i>	The family enjoyed spending time together.
	<i>The oldest son was like a father and mother to his little sister. She turned to him for help and he was there for her.</i>	During a time of crisis, the siblings tried to help each other.
	<i>One day Danny's [a fictitious name for the child with TS] little sister was punished in school and she was put in his classroom. Danny stood up and said, "My sister screws my little brother all the time, over and over". She began to cry and felt horrible, and did not want to go back to her class.</i>	Siblings were embarrassed by the behaviour of the sibling with TS.
	<i>We live well, take care of the children, work and live a "normal' life. Even when there are outbreaks and yelling, we have learned to live with it.</i>	Parents love and care for all their children in the family.
	<i>The most difficult was when his brother tried to create contact and speak to him, and he [the child with TS] would repeat the words that his brother said and would not stop.</i>	Inability to communicate with the sibling with TS led the other brother to be isolated in his room.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>It annoyed his brother and sometimes he would get angry and shut himself in his room.</i>	
	<i>On Saturdays we go out together because we found out that she likes nature and it calms her. We wait for the weekends to spend time together.</i>	Shared family activities led to much happiness.
PARENTING	<i>I am really afraid that a day will come when I won't be able to catch him and calm him down. In particular I am afraid of adolescence.</i> <i>He will grow up, continue to curse and I won't be able to control him... that's really scary to me. Now he is small, but what will happen when he grows up [tears in her eyes, almost choking while speaking]. Sometimes I think that he will become a criminal. I don't want to think about it.</i>	Fear for the child's future.
	<i>I think I am a good mother. I invest and love.</i> <i>I have difficult days. I punish. Sometimes I punished more, and today I understand better.</i>	Despite the difficulty, the mother thinks she is a good mother.
	<i>He caused me to see myself as a good and understanding individual. I am much more patient with the other family members and very active in an organisation that helps other families.</i>	The positive view of the parent.
	<i>People don't understand that he has a disorder and look at him strangely. They think he isn't disciplined.</i>	The need to stand up for the child in the environment to allow him to feel secure.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>I don't have the strength to explain it to everyone, but I know that I have to do so for his sake.</i>	
	<i>We always had to prevent him from having the opportunity to be alone and to hurt himself. We would follow him everywhere. It was as though he was hospitalised, and all the time he couldn't leave the house. We had to be with him all the time until he stopped the behaviour once and for all, and every time he had an attack, obsessions, we would give him medication, and then stop when he calmed down.</i>	There was a need for continual following and watching.
	<i>He was unable to wash himself, he could not dress himself. I had to do everything. He was not able. He was in such a state that if he spoke to me, you couldn't be sure if you were communicating with him, it was as though you were talking to him and then didn't know if he was receiving what you said. He was in his own world. At the end of the day I was weak and nervous and couldn't help the other children.</i>	Worrying about the child with TS and intensive care for him made her neglect the other children in the family.
	<i>At home he would go crazy and not stop yelling and cursing. Sometimes it would drive me crazy. I would put him in the room and hit him hard and ask him not to come out.</i>	Lack of information about the condition caused parents to act violently.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>I didn't know he couldn't control his behaviour.</i>	
	<i>After he was expelled from school in Year 9, he spoke about the fact that he had been considering suicide and all kinds of things, and I felt hopeless. How can I make him happy again? It is a feeling that I simply remember that I cried about. I cried and simply asked the doctor to help me, to find a way for my child to be happy. I had no one to turn to.</i>	Fostering for their children.
	<i>I try to keep my relationship with my husband and my other children strong, because this gives me the ability to help my child with TS.</i>	
	<i>I am really afraid that a day will come and I will not be able to catch him and calm him down. I am particularly afraid of adolescence. He will grow up, continue to swear, and I won't be able to control him. This really scares me. Now he is little, but, what is going to happen when he grows up? [Almost choking with her tears, the mother continued and said:] 'Sometimes I am afraid that he will be a criminal, I didn't want to even think about it.</i>	
	<i>Knowing that my child could not control himself because he has a neurological problem tore me apart, for years I blamed him for his behaviour and punished him.</i>	

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>Sometimes I even used physical force to control him.</i>	
	<i>I always feel that I have to speak up with teachers, doctors and specialists to make sure my child gets good care.</i>	Developing good decision making strategies, advocacy.
	<i>Attention and energy are focused too much on the child with TS, and I feel that I am not sensitive to the other children. I feel that this is not fair, but I do nothing about it.</i>	Ability to care for all their children.
	<i>I feel that I am not strong and am unable to help my children, and sometimes my response to the situation is introversion and silence.</i>	
	<i>My child with TS is a gift. He makes me feel like a good, open and understanding person.</i>	The positive perspective on having a child with TS.
EMOTIONAL WELLBEING	<i>My daughter once wanted to run away from home. She couldn't deal with him [the child with TS]. She couldn't speak to us about it. She felt tension. He bothered her. She couldn't do her homework because of the noise and yelling. He hit her and she couldn't manage and tried to run away. It was only later, when we talked about what happened that we understood how much she was suffering.</i>	Parents didn't see the suffering of the sister because they were worried about the child with TS.
	<i>The children in the family needed a lot of encouragement and I didn't give it to them. Sometime I would hear his brother say that he wanted to kill himself.</i>	Inability of parents to help and support all siblings led to much tension.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>Sometimes I would take his little sister to therapy at the psychologist because she starting hitting and biting children in kindergarten and this was new behaviour.</i>	The ability to care for the other children in the family.
	<i>My mother and father rarely visit us. They say I don't know how to raise my children. My mother in law is very old and doesn't really interfere. The financial help she provides is enough.</i>	Inability of the extended family to understand and help.
	<i>One friend always blamed me for giving medicine to my child and medicine isn't good. What she didn't understand was that I had no choice. She would make me angry with her comments and I told her to stop coming over. She made me feel guilty. What kind of a friend is that?</i>	Inability of friends to help and understand made the mother feel guilty.
	<i>My parents did not want to help because they thought I didn't know how to raise my children and they weren't able to cope.</i>	Lack of Support from family members and friends.
	<i>The feeling that the environment blames me for not knowing how to raise my child and that I am not a successful mother is very difficult, in particular, when it comes from the people closest to you.</i>	The feeling that the mother is not a good mother.
	<i>Friends stopped visiting us, for some the swearing was very difficult so they stopped coming (for a visit).</i>	Having contact with family and friends.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
FINANCIAL WELLBEING	<i>I wanted to provide therapy to the other children but it cost a lot of money, and my husband was not working. Maybe if we had taken care of them earlier, things would not have become so bad.</i>	Inability to give the other children private medical care affected the family's health.
	<i>I work here at home because I have to be available. They call from school all the time. Sometimes I have to go and bring him home. In Year 1, either I or my husband spent most of our time in school. It was really difficult.</i>	Parents needed to be available and help when necessary.
	<i>The only problem is money. As a young couple it was difficult financially, and all the therapy and psychologists cost a lot of money. We pay a lot of money to an assistant to watch him in school. My husband took on two jobs to pay for all of these expenses.</i>	Parents need to change their work to care for their child with TS.
	<i>Now we have more money. I have a nanny and she helps. This way I can continue with my day without being exhausted and I can care for my other children.</i>	A good financial situation allows the family to employ extra help, and therefore the mother has more quality time with her children.
	<i>It is hard for me, because I need to take him to the psychologist weekly, but when my other children need medical care I also take them to the doctor that is closest. I try to take care of all my children.</i>	Health care, education and transportation.
	<i>I am always busy travelling from place</i>	Searching for help

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>to place trying to find someone to help me, and forgot that I had other children at home. At night I cried and cried.</i>	meant parents forgot and neglected other children at home
	<i>With so much running around between different doctors to take care of the child with the TS, I have no energy left to take care of my other children, and I feel that I am neglecting them, and this causes me a feeling of great sorrow. I am devastated, and have no energy left any more.</i>	Families did not have the time and energy to take care of the other children and felt sorrow.
	<i>When he started to jump and made noises at the mall, I couldn't control him and people looked at him as if he was crazy.</i>	Not feeling safe in the community.
	<i>We take care of the children; we try to give them what they need. The problem is not the TS, but the responses of the environment.</i>	Responses of the society made it more difficult.
	<i>We were torn between our feeling of empathy for him [the child with TS] and the need for discipline because he was so devastated about what he had done and promised never do it again.</i>	Parents were torn between the demands of the environment and their empathy for the child with TS.
SERVICES FOR THE CHILD WITH TS AND THE FOR THE CHILD'S FAMILY	<p><i>After I discovered that my child almost stopped breathing, I decided to look for a more serious doctor who could explain what was going on with my child.</i></p> <p><i>Until now, no doctor has been able to</i></p>	Lack of assistance from medical and psychological services.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>tell us what's wrong with the child. Why hasn't anyone told us what the child has ... all these problems all these years.'</i>	
	<i>My husband is the only provider for our family so we couldn't afford to go to a private clinic. Sometimes we change the priorities in the family, but it is difficult. Our children's health is most important for us and we are doing everything we can to give them the best treatment. The doctor at the public clinic didn't diagnose the symptoms, and it was only when he [child with TS] was an adolescent and my family almost fell apart that we went to a psychiatrist on a private basis and he diagnosed the problem. As soon as we started to give him medical treatment he calmed down.</i>	The fact that the syndrome was not diagnosed earlier affected the family.
	<i>If my child was happy in school, we would be a happier family.</i>	Inability of educational services to help.
	<i>The difficult problem began in school, in Year 1. He didn't fit in, he left the classroom, yelled and cursed without end, talked and talked, didn't let the teacher teach. I knew that I had to fight for him.</i>	
	<i>I see that the child is behaving in a strange way. My husband was out of the country, and I decided to look for a more serious doctor than the one I had. I made an appointment with Dr. S. in</i>	The family does not receive answers from the public health system and must look for a diagnosis in private health care.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>Zichron Yakov (a name of a city in Israel), because until then no one had told me what was wrong with my child.</i>	
	<i>We took him to a private psychiatrist and he said we should do a CT because of the attacks but he did not give us any diagnosis.</i>	Parents felt a need for a diagnosis.
	<i>After much suffering, my friend gave me an article to read. She studies psychology and she said she thought she knew what my child has. The article exactly described the attacks and the ceremonies that he has, and then I asked the psychiatrist if this is what my son has, and he said he doesn't like to call it that name, but that is what he has. It's a shame we had to wait so long to know what my son has. I feel a relief knowing that my son has a neurological problem and that his condition has a name.</i>	The shock of not receiving answers from doctors.
	<i>Only after he began taking Respiridal and Favoxil, which really helped him, yes, he couldn't function without it, the school agreed to accept him. Before he received the medicine he would start to go to school and two months later the school would send him home.</i>	The education system conditioned the child's return to the system on taking medication.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>The big problem started in school. In Year 1 he didn't fit in, left the room yelled and cursed without end, spoke and spoke, didn't let the teacher teach. They asked us to find a different school for him and said that it did not suit them for him to remain in the school.</i>	The education system sent the child away.
	<i>He talked to me about the fact that he considered suicide and all kinds of things that he wanted to do to himself, and I felt helpless. How could I help him and give him back joy in life? When I remembered this feeling I cried. I simply asked the doctor to help me, as if he could find a way for me to give him back joy in life. The doctor didn't know how and didn't help.</i>	Medical services could not provide help to the family.
	<i>When the child reached the age of 4 we went to a psychiatrist because he had temper tantrums, he would yell, he didn't stop repeating words over and over again. The doctors diagnosed ADD and recommended Ritalin. He started to blink his eyes in a strange way and make noises.</i>	The diagnosis was not deep or professional enough.
	<i>I knew that I had to fight for him. I asked the TS Organisation in Israel to come to the school and lecture the teachers on the phenomenon.</i>	The need to advocate for the child opposite the education system.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>I turned to the Regional and Town Support Centre which is a professional centre that provides care to children with academic difficulties in the regular education system, and asked for an assistant to stay with him. If I had not gone directly to the supervisors in the education system and the support centre, the school would have thrown him out, and he'd be in special education in a school for disturbed children.</i>	
	<i>After we found an open school and he was accepted, he cursed a child and called him 'dirty Ethiopian'. The child was the son of the therapist in the school, and they called and said they could not keep him because of his cursing [with tears in her eyes]. We didn't know what to do. I knew that I had to fight for him.</i>	The education system could not cope. Difficulty coping alone when facing the education system.
DESCRIPTION OF THE BEHAVIOUR OF THE CHILD WITH TS	<i>At age 9-10 he would perform a sort of dance. He would jump and spin and you couldn't take him to the store. He would spin around all the way to the store and back home. He also had a tic that was like a dog barking and an attack. His behaviour was very severe, disruptive in school. The neighbours spoke about it. It also bothered the family. It was very annoying and so we stayed at home.</i>	The child's strange behaviour embarrassed the family and those around him spoke about it (neighbours and school).

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>At that time he had a lot of temper tantrums, cursing and yelling and unending speech. He kept saying the word 'sex'. Every sentence and every situation, he would say 'sex'. He was suspended from school and we had no one to turn to for help.</i>	Severe behaviour that is socially unacceptable, especially in the education system.
	<i>The attacks became more complex, and once he started barking and making a noise with his nose, like Snoopy. It was difficult to go out of the house with him.</i>	Family's fear of the reactions of the environment.
	<i>He didn't attend school for a whole year because of his behaviour and I was at home with him.</i>	The education system could not deal with the severe behaviour of the child with TS.
	<i>At age 10 he still wet his bed and even wet himself during the day. He would throw up - a lot. And he said he didn't want to do it and wasn't doing it on purpose. But he would throw up all the time. And once he threw up on the walls. My husband said he could not stand it anymore, and he said, it's either me or the child. So I said to him, you need a vacation. Take your things and go to Jerusalem, and then he left and came back a few months later.</i>	The father couldn't bear the child's behaviour and decided to leave and not cope.
	<i>The mornings were a nightmare. He couldn't move, wouldn't do anything. Wouldn't get dressed, wouldn't eat. He was like a VIP.</i>	The child's behaviour causes pain and a sense of helplessness for the parents.

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<i>I had to dress him and he would not cooperate. He only yelled and disrupted and cursed. By the time he left the house I was exhausted and my whole body hurt. At night he went to bed late. It was difficult for him to fall asleep. I feel helpless.</i>	
	<i>He would spit all the time, constantly, constantly. It embarrassed me and I couldn't stop it.</i>	The child's uncontrolled behaviour embarrassed his mother.
	<i>The most difficult phenomenon was his obsessive thoughts about his faeces and his sense that he has a bathroom inside. He put his hand inside, all the time thinking about it. We had to watch him and ensure that he wouldn't go to the bathroom alone, so we had to go in with him and keep him from sticking his hands inside</i>	The need to be constantly watchful.
	<i>He was a difficult toddler, crying all the time, all the time nursing, not sleeping, and constantly moving. He wore me out and I didn't understand why. I was a young mother and I didn't know if it was 'normal' or not. As a baby, he didn't sleep, moved all the time, jumped from place to place. I couldn't leave him alone for a moment because I feared he'd fall.</i>	Difficulties started when he was toddler

FQOL Domains	Family Perceptions Of Their FQOL	Tentative Interpretation
	<p><i>One psychologist recommended that I leave him alone and told me that I shouldn't tell him to stop all the time because he could not control his behaviour. But the other psychologist said we have to set limits and teach him that it isn't nice to act this way, and with time he'd learn to control himself. I was confused and needed more information and guidance. So we went to a private psychiatrist which cost much money.</i></p>	<p>Lack of appropriate information confused parents.</p>

5.4 Interview Findings according to FQOL Domains

5.4.1 Family Interaction

The first FQOL domain, *Family Interaction*, focuses on the relationships among family members (Turnbull, et al., 2004), relating to the characteristics as they emerged from this research, i.e. the variables: *Open Discussions*, *Enjoying spending Time together* and *Love and Care*.

The fact that a family has a child with TS affects family dynamics and relationships among family members. Even though positive relationships between and among family members are very necessary for a good FQOL, for some families, their relationships apparently undergo significant changes because of the pressure they experience: *'The many pressures we faced made our family life impossible'*.

For other families, dealing with the situation caused almost total dissolution of the family:

The stress of our lives tore my husband and me apart, not an uncommon story in the life of a child with TS. We could not understand and we needed guidance to go through all this. We couldn't function as a family.

When one member of the family could not function and separated him or herself from the situation, this could cause much frustration:

My husband detached himself... it was his way of coping with the new situation. He didn't want to talk and couldn't help and I felt frustrated.

And when the entire burden fell on one parent, in this case, the mother, it caused a lot of tension:

As if the family almost disintegrated. There were a lot of problems. In the house my husband and I are very close to one another, we have a very good relationship, but we were so tense and the tension was so hard on me. These were the nights that I cried and cried and my husband couldn't help.

Most of the families indicated that they needed guidance and some activities to maintain good relationships within the family:

I wanted to talk to my husband but I didn't know how to do so, I wish I could get some help.

Families expressed a need to share their feelings in order to function in daily life, as one mother said:

At first my husband and I had difficult times. We did not know what to do, and this created tension between us. But now I feel that it brought us closer. My husband also has TS. We found it out only after we understood that our son has Toilette. Now we speak about it between us a lot and it helps us and makes us closer.

One of the mothers said:

It was most difficult for me because all the time, almost all the time, it was only me who was with him [the child with TS], I slept with him on the couch, because there was a period when he tried to bite himself all the time, he would cause himself injury with his teeth, and I would perform reflexology, and that calmed him down, and I would lie with him on the couch, and I would caress him until he fell asleep. Those were the nights we slept on the couch. In the morning I was tired and could not function and take care of the other family members.

When a parent could not function, stress was apparently engendered, 'I wanted to talk to my husband but I didn't know how to do so, I wish I could get some help.'

Good cooperation between the parents seemed to help alleviate pressure:

Twice a week I go to exercise class and yoga and my husband puts the children to bed. It also helps. I am with myself and it is very important to me, and I relax from the stresses of the day.

Thus, according to the evidence it seems that when the father isolates himself, the mother apparently takes the responsibility for the care of the child with TS. Sharing feelings and open discussions apparently help to decrease stress.

Some families, however, commented that the siblings felt discomfort, shame, and/ or embarrassment regarding their sibling with TS. One mother said that:

For a very long time my son did not invite any friends home. At first I did not understand that his behaviour was due to a feeling of shame and that he was afraid his brother would act strangely or even aggressively towards his friends. When I decided to ask him about it, he told me that he was embarrassed by his brother's behaviour.

Information about TS might help the siblings deal with the situation in a better way, yet this will not necessarily remove all social difficulties. In another family, the mother said:

My son did not invite his friends to his home because he did not want his friends to see his sister, because he thought he would get teased.

In another case a brother's inability to communicate with the sibling with TS led the brother to isolate himself in his room:

The most difficult episode was when his brother tried to create contact and speak to him [the child with TS]; he would repeat the words that his brother said and would not stop. It annoyed his brother and sometimes he would get so angry that he shut himself in his room.

Sometimes children with TS embarrassed their siblings, and this was frustrating, as one mother explained:

One day Danny's [a fictitious name for the child with TS] little sister was punished in school and she was put in his classroom. Danny stood up and said, 'My sister screws my little brother all the time, over and over. She began to cry and felt horrible, and did not want to go back to her class.

Some siblings of children with TS showed concern regarding the future of their TS siblings. One of the brothers was afraid that his brother was going to die. When his mother asked him why he felt so, he answered:

Because at first you would go off and discuss with my father , my brother's situation in whispers, and no one told me anything, which caused me to think that something horrible was about to happen.

Siblings apparently do not understand that there are so many problems to deal with and it seems to be difficult for them to accept the fact that the child with TS needs more care. The parents try to take care of all their children.

And in time of crisis siblings often tried to understand and help each other:

The oldest son was like a father and mother to the little sister. She turned to him for help and he was there for her.

Some families enjoyed spending time together:

When we discovered that our child likes to be out in nature, we began to plan hikes every week. The little children had a good time and he was calm and happy. This brought the family closer together and allowed quality time with the children.

Others said that when they shared family activities it brought them much happiness:

On Saturdays we go out together because we found out that she likes nature and it calms her. We wait for the weekends to spend time together.

Thus, it seems that open communication within the family initiated and guided by the mother helped to clarify the feelings of the siblings of the child with TS, and helped the family to understand the needs of all family members.

Thus too, from the evidence gathered from the interviews it appears that there are two critical elements for FQOL of families of children with TS: The role of the mother within family interaction and provision of accurate and relevant information

And, it seems that openness, and airing and sharing of family members' feelings guided by the parents, are characteristics of family interaction, that can positively affect the FQOL of families who have a child with TS.

The data seem to indicate that relationships were altered when one of the children in a family had TS family, but on the whole, relationships remained good. Families attempted to develop and maintain good relations by participating in various activities together. In most families, the feeling was that there was an understanding and closeness, and the family members considered themselves to be a unified family that knew how to cope well in their daily life. As evidenced by the data:

We live well; we take care of the children, work and live a 'normal' life. Even when there are outbreaks and yelling, we learned to live with it.

Taking care of a child with TS was perceived as an opportunity to do good deeds and thus gain innate satisfaction from helping the child and the family.

The findings seem to indicate that in families of children with TS, in order to maintain a 'normal' family interaction, family members need to talk with each other about their feelings and the ways they are dealing with the situation.

This apparently enables them to feel competent and comfortable. They need to talk about their feelings of safety and fear, and what it takes to improve the sense of security in the family. The families' interviews revealed that when they were able to talk openly with each other this helped to prevent stress and generate options for problem solving. Moreover, the family members seem to have talked about how they envision the future for their children in general and for the child with TS in particular. Families also apparently need friendships and closeness with each other.

Thus, in sum, the evidence seems to indicate that the mother emerges as a critical figure within the family as a catalyst creating an atmosphere of openness, sharing and airing of feelings, and helping to improve the morale and mood of the family as a characteristic of family interaction affecting the perception of FQOL.

5.4.2 Parenting

The second domain of FQOL is parenting, which refers to the activities that parents do to help their children grow and develop (Turnbull, et al., 2004). Additionally, parenting includes 'fostering their children's independence', 'developing good decision making strategies' and 'ability to care for all their children'.

It is common assumption that parents expect to have a perfect, healthy child. Disappointment can be acute when one has a child with TS. Families may experience feelings of denial, anxiety, guilt, depression, or even anger when they are unable to help their child. One mother said:

After he [child with TS] was expelled from school in Year 9, he spoke about the fact that he had been considering suicide and all kinds of things, and I felt hopeless. How can I make him happy again? Whenever I remember that feeling, I cry. I cried and simply asked the doctor to help me, to find a way for my child to be happy. I had no one to turn to ... and the doctor did not know how to help.

The fact that the parents needed to watch their child all the time exhausted them:

We always had to watch him to ensure that he wouldn't go into the bathroom alone, so we had to go in with him and make sure he wouldn't put his hands in'.

We had to prevent him from having the opportunity to be alone and to hurt himself. We would follow him everywhere. It was as though he was hospitalised, and all the time he couldn't leave the house. We had to be with him all the time until he stopped the behaviour once and for all, and every time he had an attack, obsessions, we would give him medication, and then stop when he calmed down.

The need for continual supervision and worrying about the child with TS and the need for intensive care often meant that the parents neglected the other children in the family:

He was unable to wash himself, he could not dress himself. I had to do everything. He was not able. He was in such a state that if he spoke to me, you couldn't be sure if you were communicating with him, it was as though you were talking to him and then didn't know if he was receiving what you said. He was in his own world. At the end of the day I was weak and nervous and couldn't help the other children.'

Thus, parents often felt that they did not invest enough in their other children, and that they could not protect them and this caused them much sorrow:

Attention and energy are too focused on the child with TS, and I feel that I am not sensitive to the other children. I feel that this is not fair, but I do nothing about it.

Other parents feared that they might lose control of their child with TS, particularly fearing adolescence and fear for the child's future. With tears in her eyes, one mother said that:

I am really afraid that a day will come and I will not be able to catch him and calm him down. I am particularly afraid of adolescence. He will grow up, continue to swear, and I won't be able to control him. This really scares me. Now he is little, but, what is going to happen when he grows up? [Almost choking with tears, the mother continued saying:] Sometimes I am afraid that he will be a criminal, I don't want to even think about it.

One father said;

I feel that I am not strong and am unable to help my children, and sometimes my response to the situation is introversion and silence.

Families apparently need to maintain good relations in order to be strong enough and have the strength to help their other children. One mother said:

I try to maintain a strong relationship with my husband and my other children, because this gives me the ability to help my child with TS.

Lack of information about the syndrome sometimes led parents to act violently:

At home he would go crazy and not stop yelling and cursing. Sometimes it would drive me crazy. I would put him in the room and hit him hard and ask him not to come out. I didn't know he couldn't control his behaviour.

Most of the families indicated that if they had been given information about the syndrome they would know how to deal with it:

Knowing that my child could not control himself because he has a neurological problem tore me apart, for years I blamed him for his behaviour and punished him I even sometimes used physical force to control him.

It is commonly assumed that parents feel that part of their role is to protect their children and they expect others to support their children in order to improve their quality of services. Parents indicated that they had to develop good decision making strategies and advocacy to help their child with TS:

I always feel that I have to speak up with teachers, doctors and specialists to make sure my child gets good care.

They apparently needed the help of professionals to take care of their children's emotional difficulties:

I cried and simply asked the doctor to help me, to find a way for my child to be happy. I had no one to turn.'

They expressed a need to stand up for the child with TS in the environment in order to protect them from the environment:

People don't understand that he has a disorder and look at him strangely. They think he isn't disciplined. I don't have the strength to explain it to everyone, .but I know that I have to do so for his sake.

Many parents felt that their families had been blessed because they had their special child. One mother said:

Because of him I saw myself as a good and understanding individual. I am much more patient with the other family members and very active in an organisation that helps other families.

Additionally, they said that they became more kind, understanding, sympathetic and unselfish. Some of the families considered the fact that they had a child with TS as a gift from God. One mother said: *'My child with TS is a gift'*. She echoed the words of the previous mother, seeing herself as a good and understanding individual:

He makes me feel like a good, open and understanding person. I think that I am a good Mom. I invest more and I love, I have more patience even though I have difficult days. Sometimes I punish. I used to punish more, but now I am more understanding.

Thus, according to the evidence, the parents apparently find it difficult to take care of all of their children as they would like to, as an integral part of their parenting, and simultaneously to advocate for and provide intensive care for the child with TS. These feelings seem to characterise the FQOL of families who have a child with TS. Despite the difficulty of raising a child with TS, most of the parents perceive themselves as good and caring parents who have an open and accepting approach to their children.

5.4.3 Emotional Wellbeing

The third domain of FQOL investigated in this research is *Emotional Wellbeing*, which refers to the emotional or affective part of family members' and family life (Turnbull et al., 2004). Emotional wellbeing includes the characteristics: 'receiving support from family members and friends that relieves stress', and 'quality time to pursue their own interests'.

Most families seem to be forced to cope with their difficulties alone, and in most cases, do not receive any assistance from the extended family. As evidenced by the families:

My parents did not want to help because they thought I didn't know how to raise my children and they weren't able to cope.

My mother and father very rarely come to visit us. They claim that I don't know how to educate my children.

My mother in law is an elderly woman so she does not intervene with the TS'.

Friends stopped visiting us, for some the swearing was very difficult so they stopped coming (for a visit)'.

Inability of friends to help and understand also made the mother feel guilty:

One of our friends constantly accused me of giving my son too much medication, and medications are not good. What, doesn't she understand that I have no choice? She used to aggravate me with her comments, so I told her not to come any more. She made me feel guilty.

These circumstances apparently led to the isolation and exclusion of the family:

The feeling that the environment blames me for not knowing how to raise my child and the conclusion that I am not a successful mother is very difficult, in particular, when it comes from the people who are the closest to you.

Some parents felt that they neglected their children and did not provide sufficient care for the other children in the family:

Sometimes I would take the little sister to therapy at the psychologist because she starting hitting and biting children in kindergarten and this was new behaviour.

Inability of parents to help and support all the siblings led to much tension:

The children in the family needed a lot of encouragement and I didn't give it to them. Sometime I would hear the brother say that he wanted to kill himself.

Parents didn't see the pain of the other siblings because they were so worried about the child with TS:

My daughter once wanted to run away from home. She couldn't deal with him. She couldn't speak to us about it. She felt tension. He bothered her. She couldn't do her homework because of the noise and yelling. He hit her and she couldn't manage and tried to run away .Only later, when we talked about what happened we understood how much she was suffering.

Thus, the evidence seems to indicate that due to the inability of the parents to provide sufficient care for all the children and the inability of the extended family or friends to understand the family's difficulty and to assist the family members the family often feels isolated and alone.

5.4.4 Physical/Material Wellbeing

The fourth domain of FQOL investigated in this study is *Physical/Material Well-being*, including the following characteristics: 'medical and dental care can be obtained when needed', 'family members feel safe at home, work, school and in their neighbourhood' (Turnbull et al., 2004).

The evidence collected for this study seems to show that the health of the family is an important component of FQOL. A few families managed to achieve the right balance between taking good care of the extensive needs of the child with TS, such as emotional and health care and also taking care of the needs of other family members:

It is hard for me, because I need to take him to the psychologist every week, but when my other children need medical care I also take them to the doctor that is close to our home.

Other families reported that a good financial situation allowed them to employ extra help, and therefore they had more quality time with their children:

Now we have more money. I have a nanny and she helps. This way I can continue with my day without being exhausted and I can care for my other children.

Parents sometimes needed to change their jobs to care for their child with TS:

The only problem is money. As a young couple it was difficult financially, and all the therapy and psychologists cost a lot of money. We pay a lot of money to an assistant to watch him in school. My husband worked two jobs to pay for all of these expenses.

While others decided to work at home to be available and help when necessary:

I work here at home because I have to be available. They call from school all the time. Sometimes I have to go and bring him home. In Year 1, either I or my husband spent most of our time in school. It was really difficult.

On the other hand some families claimed that they did not have the time and energy to ensure that all members of the family received treatment for their medical health.

As the parents reported:

With so much running around between different doctors to take care of the child with the TS, I have no energy left for taking care of my other children, and I feel that I am neglecting them, and this causes me a feeling of great sorrow. I am devastated, and have no energy left any more.

Other families reported that they did not have the money and the ability to give the other children private professional care when necessary, and this affected the family's health:

I wanted to give therapy to the other children but it cost a lot of money, and my husband was not working. Maybe if we had taken care of them earlier, things would not have gotten so bad.

The feeling that they abandoned the siblings because of the amount of time they spent with the child with TS apparently made them sad and they blamed themselves for doing so:

I am always busy travelling from place to place trying to find someone to help me, and forgot that I have other children at home .At night I cried and cried.

Additionally it seems that families did not feel comfortable in the community; they felt confused by the behaviour of the child with TS when exposed to society:

When he [the child with TS] started to jump and made noises at the mall, I couldn't control him and people looked at him as if he was crazy.

They felt that the community did not want to understand:

We take care of the children; we try to give them what they need.

The problem is not the TS, but the responses of the environment.

As they became more familiar with the behaviour of the child with TS the feeling of shame that the environment's reaction to the child's behaviour engendered was apparently transformed into a sense of regret and concern that they were unable to control the child's aggressive behaviour. They understood that although it was important to give the child with TS a sense of safety, it was nevertheless necessary to control the problematic behaviour:

We were torn between our feeling of empathy for him (the child with TS) and the need for discipline because he was so devastated about what he had done and promised never do it again.

Thus, according to the evidence, it seems that the parents' abilities to provide adequate medical care for all their children and also be able to stand up for them in society were perceived as critical components of FQOL by families who have a child with TS.

5.4.5 Support and Services for the Child and the Family

The fifth domain of FQOL focuses on '*Services for the Child and for the Family*' and includes the characteristics: '*Medical Services*', '*Psychological Services*' and '*Educational Services*'.

Maintenance of the health of individual family members is seen as an important component of FQOL. When a family member has health problems this may impact that the functioning of the entire family. Most families reported some health concerns or that their health needs were not met. When they did not receive appropriate services from the health authorities it seems that family functioning was impaired:

My husband is the only provider for our family so we could not afford to go to a private clinic. Sometimes we change the priorities in the family, but it is difficult. Our children's health is the most important thing for us and we are doing everything we can to give them the best treatment. The doctor at the public clinic did not manage to diagnose the symptoms, and it was only when he [the child with TS] was an adolescent and my family almost fell apart that we went to a psychiatrist on a private basis and he diagnosed the problem. As soon as we started to give him medical treatment he calmed down.

Families reported that they did not trust the medical and psychiatric doctors' abilities to diagnose the problem. From their experience and in their opinion, most doctors did not know what was wrong with the child. One of the mothers said:

After I discovered that my child had almost stopped breathing, I decided to look for a more serious doctor who could explain what was going on with my child. Until then, no doctor had been able to tell us what was wrong with the child. Why hadn't anyone told us what the child has... all these problems all these years.

The family often did not receive answers from the public health system and were forced to look for a diagnosis in private health care:

I saw that the child was behaving in a strange way. My husband was out of the country, and I decided to look for a more serious doctor than the one I had. I made an appointment with Dr. S. in Zichron Yakov, because until then no one had told me what was wrong with my child.

Most of the families who were interviewed indicated that services provided to the child with TS were defective or inadequate especially during the diagnosis. Sometimes the diagnosis of the symptoms was a shock for the families:

After much suffering, my friend gave me an article to read. She studies psychology and she said she thought that she knew what my child has. The article exactly described the attacks and the ceremonies that he has, I was shocked. How come the doctor did not tell me what my child had? Then I asked the psychiatrist if this is what my son has, and he said he doesn't like to call it by that name, but that is what he has. It's a shame we had to wait so long to know what my son has. I feel a relief knowing that my son has a neurological problem and that his condition has a name.

Families turned to doctors for help in crisis situations and received no help:

He talked to me about the fact that he had considered suicide and all kinds of things that he wanted to do to himself, and I felt helpless. How can I help him and restore joy in his life? It is a feeling that I felt remembering that I cried. I simply asked the doctor to help me, as if he could find a way for me to give him back joy in life. The doctor didn't know and didn't help.

Even when parents asked for answers when the child was at an early age, they were disappointed when the diagnosis was not sufficiently deep or professional:

At age 4 we went to a psychiatrist because he had temper tantrums, he would yell, he didn't stop repeating words over and over. The doctors diagnosed ADD and recommended Ritalin.

Lack of appropriate information or contradictory information apparently confused the parents:

One psychologist recommended that I should leave him [the child with TS] alone and not tell him to stop all the time because he could not control his behaviour. But the other psychologist said we have to set limits and teach him that it isn't nice to act this way, and with time he'd learn to control himself.

Parents reported that if the diagnosis had been given to them earlier, it would have averted unnecessary stress and tension. This response was common among the families participating in the research. For example, one mother explained:

The knowledge that my child's disorder had a name and that they knew what was causing the behaviour led me to understand that both I and my son had done nothing wrong. It helped me to accept the fact that my son simply couldn't control his tics. Now I can tell people that my child has a neurological problem.

Another parent said that:

Knowing that my son's condition has a name brought some relief as we understood that there is situation here that needs to be treated, and it has nothing to do with the fact that we are or are not good parents. Knowing brought some relief.

In another family, the mother said:

When the psychiatrist said, your child has TS, I became dizzy. For years we had run from one doctor to another doctor and no one was able to explain what was wrong with my child.

The discovery (diagnosis) of TS and the acceptance of the diagnosis are difficult, and in many cases, it takes much time until the family receives assistance. The lack of appropriate services was repeatedly noted in the interviews as something that affected the family system. Another claim that was made was that if the child could receive appropriate services, daily coping would be ameliorated and less complex. Families began to disintegrate when they were not able to diagnose the child's behaviour: *'it was as if the family almost fell apart.'* The risk of family breakdown was especially noted during the initial period following diagnosis of the syndrome.

Thus, most of the families reported that the medical, psychological and psychiatric care that they received fell far below their needs. These services cost money, and the families with little means were unable to withstand the high expense. Often, the family's decision was to accept services from their medical insurance funds, but they sometimes felt that these did not satisfy their needs. In certain cases, out of despair, some families decided to privately seek services and to pay the high cost, which forced them to change their financial priorities in the household. Seven of ten of the families who were interviewed had to change their family's financial priorities in order to finance medical services

An additional deficiency in services that the families noted related to the consideration of the child with TS by the education system. Most of the families reported that the education system failed to accept their child's situation and lacked knowledge concerning the child's difficulties and the ability to cope with these difficulties:

The difficult problem began in school, in Year 1. He didn't fit in, he left the classroom, yelled and cursed without end, talked and talked, didn't let the teacher teach.

I knew that I had to fight for him.

A few families found a solution to the problem by turning to the Ministry of Education or to organisations that help families whose children have difficulties assimilating within the education system:

I asked the Israel TS Organisation to come to the school and lecture the teachers on the phenomenon. I turned to the Special Education Service and asked for a teaching assistant to be assigned to him.

The more severe the case, the more parents reported on the authorities' inability to help them, despite their appeals to different associations. The education system very often asserted that they were unable to cope with the difficulties engendered by the TS condition:

At that time, he had a lot of temper tantrums, cursing fits and yelling, and talking incessantly. He kept repeating the word 'sex'. Every sentence and in every situation, he said 'sex'... Every time the teacher couldn't cope with him, she would ask the assistant to take him out into the yard. At the end of the year, they asked him to leave the school.

Despite the fact that some of the children were diagnosed as having behavioural problems and not as 'learning disabled', it was often reported that the school decided not to deal with the situation:

He doesn't have a learning disability. When he is calm he is very smart and fits in well. Because of his attention and hyperactivity problems, he couldn't learn and the school system just gave up.

Parents reported that they could not trust the school because they would always call and ask them to come and take their child home or they would call to complain about the child: *'They call from school all the time. Sometimes I need to go and bring him home.'*

For this reason some families have to leave or change their place of work: *'I work here at home because I need to be available.'* Parents spend time at school because they worry about their child being expelled from school: *'Most of the time either I or my husband was at school. It was very difficult.'*

According to the evidence, the lack of appropriate services from the education system made it difficult for the family to function and in most cases they ask the family to solve the problem by themselves:

They asked us to find another school for him, and said that he was not suited to stay in the school. With great difficulty we found an open school and he was accepted, he cursed a child and called him 'dirty Ethiopian'. The child was the son of the therapist in the school, and they called and said they could not keep him because of his cursing [the mother began to cry]. We didn't know what to do.

Most families asked for help and felt as though they were abandoned. One of the fathers said in his interview: *'If my child would have been happy in school, we would be a happier family'.*

Families felt that they need to advocate for the child opposite the education system:

I turned to the Regional and Town Support Centre which is a professional centre that provides care to children with academic difficulties in the regular education system, and asked for an assistant to stay with him. If I had not gone directly to the supervisors in the education system and the support centre, the school would have thrown him out, and he'd be in special education in a school for disturbed children.

Thus, according to the evidence, it seems that services for the family and for the child with TS seem to be the most significant domains of FQOL for families who have children with TS, in particular services provided by the educational and the medical systems.

5.4.6 The Severity of the TS and its Impact on FQOL

It was found that the degree of severity of the syndrome influenced the FQOL of families who have a child with TS. Families who perceived that the child had a more severe case of TS, also perceived their FQOL as low. Parents felt helpless, and had a sense of shame and embarrassment when their child's strange behaviour occurred in front of other people outside the family, especially when they were in public settings such as a shopping mall or at school and usually preferred to stay at home, as one mother said:

At age 9-10 he performed something like a dance. He would jump and spin and I couldn't take him to the store. He would spin around all the way to the store and back home. He also had a tic that was like a dog barking. His behaviour was very severe and disruptive in school. The neighbours spoke about it. It also bothered the extended family. It was very annoying and so we stayed at home.

The uncontrolled behaviour caused embarrassment especially when the child with TS couldn't control his unsociable behaviour:

He would spit all the time, constantly, constantly. It embarrassed me and I couldn't stop it.

Some families seemed to be very concerned because of the reactions of the environment:

The attacks became more complex, and once he started barking and making noise with his nose, like Snoopy. It was difficult to go out of the house with him.

For some families this behaviour generated pain and helplessness, not only outside but also at home and this exhausted them:

The mornings were a nightmare. He couldn't move, wouldn't do anything. Wouldn't get dressed, wouldn't eat. He was like a VIP. I had to dress him and he would not cooperate. He only yelled and disrupted and cursed. By the time he left the house I was exhausted and my whole body hurt. At night he went to bed late. It was difficult for him to fall asleep. I feel helpless.

Some children with TS had severe and dangerous tics and obsessive behaviour that needed to be constantly watchful:

His obsessive thoughts about his faeces and that he had a bathroom inside of him. He puts his hand inside, all the time thinking about it. We had to watch him to ensure that he wouldn't go to the bathroom alone, so we had to go in with him and keep him from sticking his hands inside.

Parents felt they did not have sufficient appropriate information and advice on how to treat their child with TS especially when the situation was more difficult and they required more help.

The findings also appear to indicate that the families whose children had what they perceived as a more severe case of TS assessed the state of services as lower than families who considered that their child had a milder case of TS, especially with regard to 'support and counselling' and 'information'. It was considered more important to receive the necessary information from different professionals such as doctors and psychologists when the severity of the child with TS was perceived as more difficult. Parents turned for help but apparently they usually did not find useful responses and this frustrated them. It seems that the guidelines of professionals were often contradictory and parents were forced to decide by themselves what to do, or to search for more help from other private professionals.

In cases where the symptoms of TS were perceived as more severe it was more difficult for parents to achieve a 'normal' life, and sometimes resulted in parents splitting up. In these cases it was the father could not bear the child's behaviour and decided to leave and not to continue to cope;

At age 10 he still wet his bed and even wet himself during the day. He would throw up – a lot. And he said he didn't want to do it and wasn't doing it on purpose. But he would throw up all the time. And once he threw up on the walls. My husband said he could not stand it anymore, and he said: it's either me or the child. So I said to him, you need a vacation. Take your things and go to Jerusalem, and then he left and came back a few months later.

The pressure and burden of taking care of the family and the child with TS was perceived as the responsibility of the mother, a perception which affected the QOL of their family. In some cases parents reported that even when the child with TS was still young and had not yet been diagnosed, they were difficult to control:

He was a difficult toddler, crying all the time, all the time nursing, not sleeping, and constantly moving. He wore me out and I didn't understand why. I was a young mother and I didn't know if it was 'normal' or not.

As a baby, he didn't sleep, he moved all the time, jumped from place to place. I couldn't leave him alone for a moment because I was afraid he'd fall.

According to the parents, the most difficult aspect of their coping with the external world was the frustration involved in the child's assimilation in the education system, particularly when a child with TS had more severe symptoms. Parents were disappointed with an education system which apparently could not deal with a severe case of TS:

A whole year he wasn't in school because of his behaviour and I was at home with him.

According to the parents' perceptions, the severity of the TS affected all the FQOL domains, especially the level of services received for the child and the family. Parents spent much time and effort in search of professionals who could effectively help. They sometimes felt the need to change their place of work and to earn more in order to get a better diagnosis and treatment for the child paying substantial fees for the advice of a private medical specialist. This situation seemed to impact on the rest of the family and especially the siblings who had to give up some of their requirements. When the severity of the child's TS was perceived as more severe, parents spent more time with them, ceasing their social tasks and activities outside the house. In some cases the burden fell on one of the parents, a situation which caused a lot of stress and tension.

Thus, the evidence seems to indicate that if the symptoms of the child with TS were perceived as more severe, the family appeared to encounter greater difficulties in dealing with the situation. Parents accordingly expressed a great deal of disappointment with the education system which was found to be a dominant variable affecting the FQOL of families who have a child with TS.

5.5 Summary

In conclusion, the results of these interviews seem to indicate that parents generally expressed their satisfaction with their FQOL. They tried to do everything to maintain family unity.

Despite these efforts, it seems from the findings that the main difficulty in relations with their children apparently stemmed from a lack of understanding because brothers and sisters needed explanations regarding the medical situation that affected their sibling with TS.

This finding is supported by the questionnaire findings which indicate that of all the FQOL variables, the *Family Interaction* variable was rated lowest by parents and siblings, i.e., parents and siblings considered family interaction to be an important factor in assessing FQOL, but expressed a lack of satisfaction with the existing situation.

It seems that siblings needed to be included more in family discussions than they were, to prevent mistaken thinking about their sibling with TS, and to prevent frustration and unnecessary stress. These explanations about the nature of TS might have helped siblings in their daily coping within the family and also with their friends in school. To the best of the researcher's knowledge, no previous research has focused on the difficulties of siblings of children with TS.

Thus too, the evidence seems to show that there was a clear need for information regarding the syndrome among the parents. They complained that if they had known earlier what was wrong with their child, and if they had had a definition for the phenomenon, this would have helped them deal more effectively with the difficulties stemming from TS both within the family and with their extended family and society in general.

Another matter mentioned by parents was that due to the fact that they did not receive information that would have helped them to understand their child's condition, they had tried to discipline their child with TS, sometimes even using violence, and blaming the child for acting strangely. This had left them with guilt feelings that for so many years they had behaved inappropriately. After the diagnosis, they did not know how to compensate their child for the suffering that they had undergone.

Another issue that clearly emerged from conversations with parents was the importance of their cooperation one with the other. It seemed that in families where parents did not cooperate, where all the pressure fell on one parent (in the researched cases this was always the mother), these families did not function effectively, and everyone suffered, including siblings, because parents did not have the time and energy to care for their children's needs, and they felt neglected.

Thus, it seems that cooperation between parents is important so that the family can continue to function as a family who love and care for each other.

This finding also relates to the variable: *Family Interaction between Parents*, and is supported by the questionnaire finding, according to which parents considered family interaction to be an important and very influential factor for FQOL, but expressed their lack of satisfaction with the level of family interaction.

Thus, the evidence seems to indicate that the family needs to receive correct and appropriate information about the syndrome, and to share the information with their children, this was a dominant variable for FQOL. To achieve this, parents apparently needed to cooperate effectively in order to allow the family to lead a 'normal' life and enjoy a good level of FQOL.

Another issue highlighted in the interviews was that parents felt they needed to advocate for their children. They apparently felt a need to speak on behalf of their child and to explain the child's condition and needs to the education system and to society.

This finding is supported by the questionnaire findings which indicate that parents of children with TS considered that it was very important to receive the services provided to them by the medical system, psychological system and counselling system, but were disappointed and dissatisfied with these services. They explained that, they had to do the work themselves. They needed to explain the syndrome and its ramifications to the education system and to the extended family and friends. The feeling was that if someone could assist them and explain the child's condition and its implications, at least to the education system, they would not have to do this work themselves, and it would alleviate much pressure from them, allowing them to spend quality time with their children.

They felt that if they did not speak on their child's behalf and explain the nature of the child's condition to the environment, particularly the school system, daily coping would be much more difficult. Therefore, they did this both for their child and for themselves. What disturbed them most was the lack of ability of the extended family and friends to understand TS, and the fact that their child was not doing what he did because of any lack of parental control or lack of discipline. They had difficult guilt feelings when the community communicated a sense that they were not good parents.

Thus, the environment's attitude apparently caused families to feel isolated.

The questionnaire findings indicated that mothers and fathers were dissatisfied with support received from the extended family and friends; despite the fact they considered that this support was very important.

There was a significant difference between the perceptions of services and support by families who perceived that their children had severe symptoms of TS in contrast to such perceptions by families who perceived that their child had milder symptoms.

These findings are supported by the questionnaire findings, which show that the severity of the syndrome influences the FQOL of families who have a child with TS especially in the education system.

Although the severity of the TS affected all areas of FQOL, the difficulties involved in assimilation within the education system were most critical for the family. It was reported that the education system's inability to cope with these difficulties often led to the child with TS remaining at home without an educational framework. This situation was a primary cause for the feeling of stress and imbalance in the family.

Thus, the interview evidence seems to indicate that in all the families, the education system constituted their main difficulty, and the most critical domain for families' FQOL, and this engendered the parents' anger and frustration.

The education system appears, in the present research, to be the most influential factor for FQOL. Parents argued that the education system was not interested in coping with children with TS, and apparently does not have the necessary tools. The easiest solution was to renege on their responsibility for the children and remove them from the system. Parents changed their priorities at home and changed their work places to be more available when called from school. This was done to enable the child to remain in school. This caused much tension and often damaged their ability to function as a family.

Thus, the parents' testimony seems to indicate that if the child was happy in school and the system knew how to cope with him, their FQOL would be better. As expressed by one of the fathers: *'If my child would have been happy in school, we would be a happier family.'*

The statements relating to services provided to the child with TS in the questionnaire all received low ratings, i.e., the parents considered *Services for the Child* to be very important, but expressed disappointment and lack of satisfaction with these services. The greatest disappointment was generally with the psychological services. Although *Special Education Services* received a low rating on the questionnaires indicating a lack of satisfaction with the service, this finding was not significant.

Only the interview findings emphasised parents' frustration and great disappointment regarding the deficiencies of the education system. It is possible that the questionnaire questions relating to the education system did not satisfactorily allow such expression.

Thus, from this limited piece of research the data suggests that, on the whole, parents did not perceive that the child with TS created problems in the family nor did the child seriously affect the FQOL. Rather it was the reactions of extended family, friends, and in particular the education system which had the strongest impact on FQOL.

This finding appears to be very important, and constitutes a very strong statement from the families, namely, that the family will manage as a 'normal' family if the necessary help is provided.

Thus, it seems that it is not TS itself that is perceived as the problem affecting the FQOL but rather the lack of means to ensure the effective functioning of the family.

5.6 Integration between the Findings emerging from the Interviews and the Questionnaire

The following points summarise the results of triangulation of findings from the interview and findings from the questionnaire

1. The need for parental cooperation: A lack of understanding that siblings needed explanations regarding their sibling with TS created a need for improved cooperation and communication between the parents and between parents and children. It seems from the research findings that families, which did not cooperate, did not function well and everyone suffered, including siblings, because parents did not have time and energy to care for the needs of their children, who consequently felt neglected.

This finding was supported by the questionnaire finding, that parents considered family interaction to be important, but expressed their dissatisfaction with it.

2. The need for information: Parents needed clear information regarding TS and its implications. Lack of information caused them to act inappropriately and when they later understood their misunderstanding they regretted the way that they had related to the child previously and expressed guilt feelings that for so many years they had tried to discipline their child with TS, sometimes using violence, and blamed the child for acting strangely. This information was perceived by them as an important tool that helped them deal within the nuclear family and with their extended family and society in general in a better way.

This finding is supported by two findings from the questionnaire: the families' lack of satisfaction with support received from the extended family and friends; and the families' lack of satisfaction with medical and psychological services, which should assist and provide appropriate and correct information.

Parents considered that support from family and friends was very important and also considered special psychological services to be important. Parents expressed a lack of satisfaction, particularly with psychological and counselling services, which should have provided information on the syndrome, but did not.

On the other hand, this finding contradicts the finding emerging from questionnaire, regarding quality of parenting. Parents gave a high rating to both the importance of parenting and their level of satisfaction with their parenting, i.e., they did not consider themselves to be unsuccessful parents although they had blamed the child for his behaviour for years until they understood that the child had a recognised medical difficulty and needed help. Apparently, the parents attributed the responsibility for their violent behaviour towards their child to the various services which did not provide answers and explanations regarding the syndrome, and apportioned less blame to themselves. It is also possible that today they consider themselves to be good parents and their behaviour that they showed towards the child before the diagnoses is simply perceived as irrelevant because it was in the past.

3. The need for explanations: It seems that the main difficulty in relations between the parents and their children apparently stemmed from a lack of understanding that siblings needed explanations regarding their sibling with TS. Thus, provision of information emerged as a significant component in families who had a child with TS.

This finding is supported by the questionnaire findings which indicated a disparity in rating between importance of *Family Interaction* and satisfaction with *Family Interaction*, based on parent's and siblings' ratings. All the respondents, parents and siblings, expressed a lack of satisfaction with family interaction, although they considered family interaction to be an important factor affecting their FQOL.

4. The need for advocacy: Advocacy emerged as a significant component of FQOL of families who had a child with TS. The attitudes of teachers, head teachers, educational administrators and other non-specified people in the wider society, caused families to feel isolated.

This finding is supported by the questionnaire findings which indicated that services provided to the family, by public bodies, the extended family and friends were considered very important by the families who had a child with TS, although expressed they a high and significant degree of dissatisfaction with these services. The result was that the parents felt a need to advocate for their children's interests opposite official bodies. The need for effective **Advocacy** as a component of FQOL is therefore advanced as a unique finding that emerged from this study.

Parents felt a need to advocate on behalf of their child and to explain the child's condition to the teachers at school and to people around them. Parents claimed that if the child was happy in school and the system knew how to cope with him, their FQOL would be better.

Services for the Family received from the education system were rated as low by parents, i.e. they saw services provided by the education system as very important but expressed low satisfaction regarding these services. This finding was supported during the interviews in which the education system was perceived to be the most dominant factor affecting FQOL especially when the child had a severe case of TS. This deficiency created the need to advocate for the child with TS before the teachers and other support providers in the education system.

6. Influence of the perceived severity: The perceived severity of the syndrome emerged as a critical component in FQOL of families having a child with TS. Parents perceived their QOL as more difficult/ OR more problematic when the child with TS was perceived as having more severe symptoms.

This finding is supported by the questionnaire findings which indicated that the severity of TS influences all the FQOL domains of families who had a

child with TS, especially in relation to financial abilities of the family to acquire services for the child and family.

Thus, when the severity of the syndrome, was perceived as stronger, a greater need for support and help was expressed. It seems that the extended family and friends have strong impact on the FQOL of the studied families. However, of all QOL domains the *Education System* had the strongest impact on FQOL. In addition, the importance of all QOL domains is strengthened as the severity of the syndrome increases.

5.7 Summary

This chapter presented the research findings which form the basis for the discussion and interpretations of the integrated findings that emerged from this study presented in the next chapter, Chapter 6.

Chapter 6: Discussion and Interpretation

6.1 Introduction

Chapter 6 develops a narrative, by interpreting and evaluating the findings, and produces an integration of the main results to form the foundation for a new view of FQOL of families who have a child with TS. The present chapter indicates how the interpretations and evaluations are supported by the theoretical perspectives, which underpinned the conceptual framework of this research and indicates relations between the findings and other relevant literature.

Thus, the chapters reiterate the research questions including the aims of the research; offer an overview of the results, and then interpret the findings that support existing knowledge. Finally, the chapter suggests a new interpretation for the recognised sub-domains of QOL which influence the FQOL of families with a child who has TS, and establishes arguments towards their introduction in the following chapter, Chapter 7: Conclusions.

The objective of this research was to study the perceptions of families of children with TS in Israel regarding their FQOL. The research aimed to highlight the FQOL domains which might be affected by raising a child with TS in the family, with the purpose of indicating directions for future research and therapeutic efforts to help these families improve their FQOL. Therefore, the main research questions addressed in this study were:

1. Which FQOL domains are perceived by family members to be affected by their child with TS?
2. How do different families raising a child with TS perceive their FQOL?

This chapter presents a discussion of the integrated findings as they emerged from the various research tools. The findings indicated that the following components were important and influential for the FQOL of families raising a child with TS:

6.1.1 The need for Co-operation between the Parents emerged as a significant component for families who have a child with TS

Co-operation and good communication between parents helps all families to function more effectively. However, in the context of children with TS it appears to have increased significance. It was found that the 'need for co-operation between the parents' was perceived by family members as one of FQOL variables that affects FQOL. The research showed that when families did not cooperate, they could not effectively and successfully manage their in daily life, so that family members endured a feeling of distress, including siblings.

This finding is supported by the findings from the questionnaires distributed to the families (parents and siblings), which indicate that rating of the '*family interaction*' variable, that includes 'family routines', 'relationships among family members and 'communication' was low. It seems that most families thought that "family interaction" was very important but expressed their dissatisfaction with the existing interaction within their families. Mothers tended to give lower rating for "family interaction" than the other family members, while siblings gave higher ratings for this variable.

This finding is supported by the study of Wang et al. (2006), who examined FQOL among families with children with mental disabilities, and found a difference between mothers and fathers in their ratings of 'family interaction', with mothers tended to give lower ratings to 'family interaction'. It may be that mothers considered 'family interaction' to be more important because in this study it seemed that they were the ones who accepted the role and overall responsibility for the family. They were in continual contact with educational and care-giving entities; they were the ones who accepted responsibility for siblings and for all other family members. As one mother said:

It was most difficult for me because all the time, almost all the time, it was only me who was with him (the child with TS).

This role apparently engendered pressure and feelings of stress. Cooperation between family members could help mothers especially in their daily management of the family and in particular might relieve stress (Wang et al., 2004).

The conjugal relationship was also found to have a most powerful influence on parent-child relationships. It was found that the quality of interaction between the parents and their ability to cooperate, providing support and help to one another in caring for the other family members was an important component of their FQOL, because this affected the ability of the family to cope more effectively with their daily struggles:

Twice a week I go to exercise class and yoga and my husband puts the children to bed. It also helps. I am by myself and it is very important for me, and I relax from the stresses of the day.

The evidence also seemed to indicate that, open discussions and cooperation between parents helped to alleviate their pressure. As one mother expressed:

I feel that it brought us closer. My husband also has TS. We found it out only after we understood that our son has TS. Now we speak about it between us a lot and it helps us and brings us closer.

This finding is supported by the literature. According to Turnbull & Turnbull (1990) and Wang et al (2004), open communication among family members is an important way to relieve stress especially when there is a child with a disability at home. This research showed that parents needed to talk to each other in order to understand the family's difficulties and facilitate their problem-solving, but they often did not know how to start the conversation and needed help in doing so. They knew that open discussions could help them deal with the situation in a better way:

I wanted to talk to my husband but I didn't know how to do so, I wish I could get some help.'

Moreover, it was found that when one family member did not cooperate and was unable to deal with the situation of living with a child with TS in the home, this affected all other family members .As one mother indicated:

The stress of our lives tore my husband and me apart We needed guidance to go through it all. My husband detached himself ... it was his way of coping with the new situation. He didn't want to talk and couldn't help, and I felt frustrated. We couldn't function as a family.

Thus, the lack of cooperation between parents, due to one spouse's (in these cases the father) inability to face the difficulty and cope, caused much tension. As one mother said:

We were so tense and the tension was so hard on me....almost the entire time I was with him [the child with TS] ... and my husband couldn't help.

Apparently, this stress both affects and is affected by the quality of the relationship and communication between the parents and in turn affects the quality of communication between them and their children. This finding is supported by studies which have found that depressed parents of children with disabilities, display less affection and happy emotions, have more problems in communicating and are characterised by less unity and more conflict in the family than do non-depressed parents (Weizman et al.,1992).

In addition, it was reported that siblings often felt neglected, especially when one parent was unable to handle the situation and the entire burden fell the other parent. As noted sadly by one mother:

At the end of the day I was weak and nervous and couldn't help the other children ... the children in the family needed a lot of encouragement and I didn't give it to them. Sometimes I would hear his brother [the brother of the child with TS] saying that he wanted to kill himself.

The parents' feeling of inability to help and support all the siblings apparently led to much tension and feelings of despair.

The research found that mothers generally handled all daily issues alone, such as going to the doctor or the psychiatrist to look for better diagnosis:

I decided to look for a more serious doctor than the one I had....because until now no one told me what was wrong with my child.

In general, it seems that it was the mother who had the greatest responsibilities toward the child with TS.

They call from school all the time. Sometimes I have to go and bring him home.

Mothers ran from place to place to find appropriate solutions for the child. As one mother explained:

with so much running around between different doctors to take care of the child with the TS, I have no energy left for taking care of my other children, and I feel that I am neglecting them, and this causes me a feeling of great sorrow. I am devastated, and have no energy left any more.

Due to this situation, mothers became exhausted and lost their patience and found themselves neglecting the needs of the other children in the family.

In addition to the academic difficulties, children with TS need regular psychological or psychiatric care. Therefore, parents felt that they needed unending time to deal with the difficulties of the child with TS. As a result, there is little time left for the other children in the family, and siblings again feel neglected:

Attention and energy are too focused on the child with TS, and I feel that I am not sensitive to the other children. I feel that this is not fair, but I do nothing about it.

As a result, parents sensed feelings of guilt and remorse. Despite the knowledge that they were neglecting their other children, some parents did not change their behaviour, claiming that they were unable to stop therapy for the child with TS and were unable to provide a better response for the other children. Running back and forth to treatments exhausted them and sometimes they almost forgot that there were other children at home. A mother said these words with tears in her eyes:

I am always busy travelling from place to place trying to find someone to help me and forgot I have other children at home. At night, I cried and cried.

Parents indicated that they were left with a feeling of incompetence because they failed to perform their parenting roles as they would have liked to.

However, this was not the case in all families. Some families realised that in order to maintain 'normal' life they had to divide their time so that they could respond to the needs of the child with TS and the needs of their other children:

It is hard for me because I need to take him to the psychologist every week, but when my other children need medical care I also take them to the doctor that is close to our home.

It seems that when parents are aware of their other children's needs, they feel more confident that they are fulfilling their parental roles as necessary.

In conclusion, it seems that in order to maintain a 'normal' life parents need to communicate and support each other. When the burden falls on only one parent, the other parent does not have the ability to manage alone. A lack of effective communication between the parents apparently generates a feeling of loneliness and isolation and parents feel that they have no one to turn to. This seems to lead to stress, which prevents them from creating a caring and understanding connection with the other family members, and affects FQOL (Wang et al., 2004).

Parents who feel tension and are unable to cooperate with one another apparently create an atmosphere of tension in the home, which is mainly expressed in an inability to openly speak with their children. Open communication among family members has been found to be one of the most important ways to relieve stress in childhood (Turnbull & Turnbull, 1990), and one of the most powerful predictors of positive sibling adjustment when growing up with a brother or sister with a disability (Gold, 1993). In the present study, it was clear that the constant need to take care of the child with TS exhausted the parents and they did not have enough time to spend with the rest of the children in the family. As a result, siblings apparently felt neglected and this in turn further frustrated the parents:

At the end of the day I was weak and nervous and couldn't help the other children.

Additionally, parents perceived themselves as unsuccessful and explained that, to their regret, their concern and attention for the child with TS, often meant that they neglected their other children. Even though they were aware that they neglected their parental duties for the other siblings, they did nothing to change the situation:

Attention and energy are too focused on the child with TS, and I feel that I am not sensitive to the other children. I feel that this is not fair, but I do nothing about it.

When parents gave more attention to the child with TS than to their 'normal' siblings; the siblings experienced the tension and stress prevailing in the family (McHale & Pawletko, 1992). Although not much research has been done to examine the emotional wellbeing of parents of children with disabilities, particularly families of children with TS, research indicates that parents do not know how to divide their time between all the children and spend more time caring for the children with disabilities. It has also been found that parents of children with disabilities are less integrated in society and do not have quality time for themselves (Gallimore et al., 1989).

Direct communication helps families to gain sensible meanings about their internal family relationships; and about their family's relations with the community and society and they can use this understanding to lower stress (Turnbull & Turnbull, 1990; Patterson & Garwick, 1994).

Conclusion: It seems that when parents supported one another and the burden of caring for the child with TS and the rest of the family was shared by the parents, the family's ability to deal with the situation was better and this helped to lower stress (Schalock, 2000, Brown et al., 2003).

It is also concluded: siblings are apparently not harmed by the mere existence of the child with TS in the family, but if parents ineffectively deal with the situation, an atmosphere of tension and stress ensues with severe negative consequences for all family members.

Since, to the best of the researcher's knowledge, there is no literature regarding FQOL of families having a child with TS, these conclusions are probably unique to the present study and contribute new understanding to extant knowledge.

6.1.2 Parents' need for clear information on TS emerged as a significant component for families who have a child with TS

The evidence gathered for this research suggests that the 'need for information' about TS is a critical component in FQOL of families who have a child with TS. As stated previously, one of the main difficulties in relations between the parents and their children apparently stemmed from a lack of understanding that siblings needed explanations regarding their sibling with TS. Broad and reliable information about the syndrome is very important so that parents can fulfil their role appropriately. Being a parent of a child with TS means facing challenges, because a child with TS is unique and special, and needs parents who will love, teach and encourage him. The child's sensitive situation is expressed in difficulty in controlling bodily movements and vocalisations, accompanied by various academic, emotional and behavioural difficulties.

In this research parents of children with TS reported that they had experienced mixed feelings of sadness, anger, guilt and shock during the period when the syndrome was diagnosed. Lacking accurate information on the syndrome, for so many years they tried to discipline their child with TS, sometimes using violence, and blamed the child for acting strangely. Once the syndrome was explained to them they felt guilt and remorse concerning the way that they had previously behaved towards the child with TS. As one father said:

'At home he would go crazy and not stop yelling and cursing. Sometimes it would drive me crazy. I would put him in the room and hit him hard and ask him not to come out. I didn't know he couldn't control his behaviour.'

Therefore lack of information about this complex syndrome, caused parents to act inappropriately, they punished, yelled and even sometimes hit their child out of feelings of despair and helplessness. Another father said:

'For years I blamed him for his behaviour and punished him. I even sometimes used physical force to control him.'

They had not understood that their child with TS could not control what he was doing and no method of coercion would help. They wanted to do their job as parents and 'discipline' the child, and once they understood the symptoms of the syndrome they regretted their previous behaviour and changed their attitude towards the child with TS. For some parents the guilt feelings engendered by the diagnosis of the child, urged them to compensate the child for their previous violent behaviour:

'Knowing that my child could not control himself because he has a neurological problem tore me apart.'

Conclusion: The provision of correct information apparently prevented unfair behaviour towards the child with TS.

Despite the feelings of guilt, parents also felt a certain relief knowing that their child has been diagnosed with TS. As one father said:

I feel a relief knowing that my son has a neurological problem and that his condition has a name.

The diagnosis brought some comfort because they understood that the child had a problem and could not control the tics or abnormal behaviour.

Conclusion: It seems that parents needed clear information regarding their child with TS, in order to deal within the nuclear family and with their extended family, friends and people around them in a more effective and appropriate way.

It was found in this research that siblings of children with TS often felt isolated for a long period of time and they needed their parents to share information with them:

For a very long time my son did not invite any friends home. At first I did not understand that his behaviour was because of a feeling of shame and that he was afraid his brother would act strangely or even aggressively towards his friends'. 'When I decided to ask him about it, he told me that he was embarrassed by his brother's behaviour.

My son did not invite his friends to our home because he did not want his friends to see his sister, because he thought he would get teased.

I thought that my brother was going to die because at first you would go off and discuss, my brother's situation with my father in whispers, and no one told me anything, which caused me to think that something horrible was about to happen.

As mentioned above, open conversations hold the potential to alleviate tension, confusion and instability within the family. It was found that parents needed clear information regarding the TS in order to be able to explain the syndrome to their children.

Parents often only understood how important information about the syndrome was, after they went through the diagnosis process and began asking questions. They realised that this information could have helped them deal within the family and with their extended family and society in general in a better way.

Another issue of concern for the parents was the need to plan for the future and prepare the child with TS for independent life. They felt that they had no control over the child's behaviour and were worried that this lack of control would lead to a lack of boundaries, and even feared that in the future this would lead the child to criminal behaviour. One of the mothers expressed her fears regarding adolescence. She argued that now at a young age she could still stop him physically and control his outbursts, but feared what would happen during adolescence. Her biggest fear was that he would physically hurt her and said with an expression of fear in her eyes:

I am particularly afraid of adolescence. He will grow up, continue to swear, and I won't be able to control him. This really scares me.

Almost choking with tears, the mother continued saying that:

Sometimes I am afraid that he will be a criminal, I don't even want to think about it.

Apparently, it seems that parents needed information about their child's behaviour from doctors and psychiatrists specialising in TS in order to feel safe and in control. They waited a long time to get answers and felt disappointment when doctors did not know how to diagnose their child, so that for many years, parents felt helpless, as one mother said:

After I discovered that my child almost stopped breathing, I decided to look for a more serious doctor who could explain what was going on with my child. Until now, no doctor has been able to tell us what's wrong with him. Why hasn't anyone told us what the child has... all these problems all these years.

It was only after they took the initiative and went independently to private specialist physicians that they began to receive helpful answers. Families cannot always finance private medical treatment, and therefore changes in family financial priorities have to be made to finance such advice and treatment. Families expressed a strong need for counselling by professionals to help them find information sources regarding the syndrome. Sources of information needed included not only information regarding the syndrome but also legal information regarding the parents' rights.

Families reported that they found it especially helpful to have partnerships with professionals, who could help them to become stronger, more motivated, with more knowledge about the syndrome.

Conclusion: It seems that information about the syndrome from professionals enables families to balance the demands of family time and develop open discussion and more appropriate ways to cope with the phenomenon of TS and consequently improves their FQOL.

6.1.3 Explanation concerning TS emerged as a significant component for families who have a child with TS

It seems that the ineffective relations between the parents and their children stemmed from a lack of understanding. The parents needed clear information in order to explain the difficulties arising from TS to the child's siblings.

Siblings were not interviewed and therefore there is not sufficient information regarding their perceptions of the quality of 'family interaction'. The impression described here is mainly derived from parents' reports. Of all the factors that affected the interaction between parents and their children, it is the quality of communication that was expressed, in particular the level of information and explanation that siblings received regarding the TS that is the most influential. A lack of comprehensive and correct explanation regarding the syndrome led to much stress among siblings, and even led, in certain cases, to incorrect behaviour on the part of siblings.

Siblings were embarrassed by the behaviour of their sibling with TS at school as one mother described:

Danny [the sibling with TS] stood up and said, 'My sister screws my little brother all the time, over and over.' She began to cry and felt horrible, and did not want to go back to her class

This behaviour of the child with TS led to social distancing. In addition, siblings did not invite their friends to their home out of fear that their brother with TS would embarrass them:

for a very long time my son did not invite any friends home ... when I decided to ask him about it, he told me that he was embarrassed by his brother's behaviour.

Another brother did not invite his friends to his home because:

He did not want his friends to see his sister, because he thought she would get teased.

In other cases, siblings were stressed because they were afraid their brother with TS was going to die: as one mother said her son had told her:

I thought that my brother was going to die because at first you would go off and discuss, my brother's situation with my father, in whispers, and no one told me anything, which caused me to think that something horrible was about to happen.

It seems that siblings of children with TS need true and full explanation from their parents about the syndrome, not only so that they can understand but also so that they can feel involved. A feeling of involvement and partnership is most critical to siblings and their QOL. Transfer of true and correct information can prevent feelings of helplessness and isolation. According to the findings, due to a lack of knowledge, siblings often created a distorted picture of the syndrome.

Obviously parents have a very important influence on siblings of child with TS and those siblings are sensitive to and affected by their parents' behaviour. Parental attitude was found to be a powerful predictor affecting sibling acceptance of a child with a disability (Powell & Gallagher, 1993). Parents who do not openly discuss the situation with all their children may generate depression among family members, and siblings may feel that their family is not a helpful resource in dealing with the ensuing stress. This may lead to feelings of isolation and feelings that it is impossible to communicate.

Conclusion: Providing siblings of children with TS with honest and accurate explanations regarding TS, and providing an open atmosphere that enables sharing of feelings guided by the parents, seems to have a positive influence on the siblings' QOL.

Honest and open communication and explanation was found to provide assurance and comfort to siblings so that they could cope with their feelings of guilt, anger, resentment, and jealousy. This finding is supported by the research of Turnbull and Turnbull, (1990) who found that the most powerful predictor affecting sibling acceptance of a child with disability was found to be parental information, in particular, the information siblings obtain from their mother. Research literature also indicates that a positive and accepting parental view was positively correlated with improved personal changes in 'normal' siblings of disabled children (Powell & Gallagher, 1993; Gibbs, 1993), as well as improved relationships with their siblings with disabilities (McHale & Gamble, 1989). It seems that the fact that a child has a sibling with TS does not necessarily impact the sibling in a negative way. There is evidence in the literature that sibling relationships involving a child with a disability may be more positive than those between comparison siblings who have no disability. Siblings have more difficulties when children's disabilities are more severe or include social deficits (Heller et al., 1999; Cuskelly & Gunn, 2003).

Conclusion: It seems that parents who had more knowledge about their child's disability were better able to provide correct and necessary information and explanation regarding the sibling with the disability and were also more successful in encouraging positive coping skills in their non-children with disabilities. This conclusion is in line with the findings of McHale & Gamble (1989) that siblings need accurate information to cope better with the situation.

6.1.4 Advocacy emerged as a significant component for families who have a child with TS

Research shows that parents of children with disabilities need support from the environment, in particular, emotional support, which is a result of understanding and empathy (Wang et al., 2004). The close environment is usually ready to support and help such families; in particular on an emotional level (Brown et al., 2003), but when it comes to the family of children with TS, the environment is not only unwilling to support and help, but even tends to blame parents for not fulfilling their roles as parents. The explanation for this attitude is that the extended family and community think that the child's irregular behaviour is the result of the parents' inability to control their child's behaviour, and they criticise them because they think that instead of disciplining, they give their child medication:

One of our friends constantly accused me of giving my son too much medication, and medications are not good. What, doesn't she understand that I have no choice? She used to aggravate me with her comments, so I told her not to come any more. She made me feel guilty.

Parents felt hurt by these responses and therefore preferred not to share their problem or ask for help, the result being that the family managed alone. Support from friends and the extended family could help the family with its daily coping with the child with TS and also help in caring for the other children in the family. In addition to the fact that these parents of children with TS did not receive support from their environment, extended family and friends, they were also isolated by their environment, which could not understand or accept the symptoms of the syndrome, and blamed the parents for not knowing how to discipline their child: *'my mother and father visit rarely. They say I don't know how to raise my children'.*

Parents found it very difficult to accept these reactions, especially when it came from people close to them, i.e., grandparents:

My parents did not want to help because they thought I didn't know how to raise my children and they weren't able to cope..... The feeling that the environment blames me for not knowing how to raise my child and that I am not a successful mother is very difficult, in particular, when it comes from the people who are closest to you.

In order for the child with TS to feel safe in their natural environment and within society, parents felt that they needed to explain the symptoms of the syndrome and they expected people in the close and more distant environments to understand without judging:

When he (the child with TS) started to jump and made noises at the mall, I couldn't control him and people looked at him as if he was crazy; we take care of the children, we try to give them what they need. The problem is not the TS, but the responses of the environment.

The main difficulty was expressed, when they needed to control the behaviour of the child with TS, and on the other hand, to allow him a feeling of security both at home and outside the home. This task was not always accomplished, because parents were often unable to control the child's behaviour, in particular outside the home.

Sometimes during these attempts parents felt that they were split between the knowledge that their child could not control what he was doing, and the need to respond to social pressure to 'discipline and control' their child:

We were torn between our feeling of empathy for him (the child with TS) and the need for disciplining because he was so devastated about what he had done and promised never to do it again.

A related issue that concerned parents was the environment's lack of understanding of the syndrome. Therefore, parents see a strong need to advocate for their children in various frameworks: schools, with friends, within the extended family, and within society in general. Advocating meant speaking up and clarifying the situation for teachers, doctors, and specialists to make sure that your child with TS received the best possible care and understanding. Parents felt that in order to be good advocates they needed clear, full and applicable information about TS. They felt that this information could help them to stand up for their child and engage with the system and the people around them, extended family and friends for the child's benefit.

The research found that parents of children with TS who participated in this study considered advocacy to be an important component of their FQOL. They thought their advocacy activities would improve services for their child:

I always feel that I have to speak up with teachers, doctors and specialists to make sure my child gets good care.

They perceived advocacy as necessary so that their child could access services and to ensure that services were improved and appropriate for the child's needs so that their child could achieve appropriate results, especially in the educational system.

The educational system was perceived by parents as the service that had the strongest influence on their FQOL. This was expressed mainly in the fact that the system does not know how to contain such children, and in most cases, preferred not to cope at all, and remove children out of the

system, causing the parents and the family much suffering as one father said: *'If my child had been happy in school, we would be a happier family.'*

Conclusion: it seems that if the educational system could be more sensitive and supportive, it would help families and thus increase their FQOL.

The findings indicated that at a certain stage of the educational process, families of children with TS met with a lack of professional response, and generally the education system preferred to remove their children from school arguing that they were not suitable for the school and did not fit in, and that it was therefore preferable to find a different solution for the child's education:

His difficult problem began in school, in Year 1. He didn't fit in, he left the classroom, yelled and cursed without end, talked and talked, didn't let the teacher teach. I knew that I had to fight for him.

Therefore, families were forced to search for other educational frames that would be amenable to their children's requirements. In most cases they had to conduct this search on their own, without any help from any authorised educational entity, and without appropriate advice:

I asked the TS Organisation in Israel (TSOI) to come to the school and lecture the teachers on the phenomenon. I turned to the Regional and Town Support Centre which is a professional centre that provides care to children with academic difficulties in the regular educational system, and asked for an assistant to stay with him. If I had not gone directly to the supervisors in the educational system and the support centre, the school would have thrown him out, and he'd be in special education in a school for disturbed children.

Conclusion: it seems that parents remained alone with the problem of their child's education and upbringing, with no help from authorities.

Some of the children with TS had to remain at home for considerable periods of time, without any educational placement burdening the family. As a result, the families became stressed and felt helpless. Many battles ensued until they managed to find a somewhat suitable educational framework, and sometimes their children had to pass through a number of different schools:

After we found an open school and he was accepted, he cursed a child and called him 'dirty Ethiopian'. The child was the son of the therapist in the school, and they called and said they could not keep him because of his cursing [with tears in her eyes]. We didn't know what to do. I knew that I had to fight for him.

The need for advocacy was especially strong when the educational system was unable to help the child to assimilate within the schools and the parents became concerned that the child would be unable to complete his education like all other children?

As mentioned in the review of the literature on this issue, in addition to vocal and motor tics, children with TS, often have educational difficulties, expressed in attention disorders, LD and other behavioural difficulties. These disturbances make it difficult for them to integrate in the educational system. After diagnosis and after parents understand their child's condition and what the difficulties are, they begin to worry about the child's academic future. Many questions arise regarding an appropriate framework, and usually, the parents must choose between a regular school and a special educational framework. Most children with TS have a 'normal' level of intelligence and in certain cases even above average intelligence (Kushner, 1999), so that parents want their children to learn in regular schools, since they have the potential to achieve good academic results and ensure their academic future and a career. However, the regular system is not prepared to absorb children with TS because of their 'strange' behaviour, and they prefer to transfer them to the special education system.

However, the parents felt that the special education system was not suitable for children with TS. This feeling is supported by the Ministry of Education, Circular of the General Manager (2009), which fails to include children with TS within the criteria for the special education population, and this fact further complicates the difficulty of finding a suitable educational framework for children with TS. Generally, children with TS are more suitable for learning in regular schools in mainstream programmes. Present policy in Israel aims to provide a response for children with academic difficulties within the mainstream programmes of the regular school system. The Israeli Ministry of Education allocates a certain number of hours to each school so that the school can provide individual or group responses to children who have difficulties, without removing them from the regular system.

By law children who have been diagnosed with ADD or other LD are entitled to additional supplementary hours in regular schools are children (Ministry of Education, General Manager's Circular, 2009). Since children with TS, usually have ADD or other LD they are included in this service. However, behaviourally, it is difficult for them to be integrated in the regular system and therefore they fall between the cracks, and parents have difficulty finding suitable frameworks for them. According to the findings of this study, some parents fight and manage to keep their children in the regular framework; others give up and move them to special education.

The parents felt that if there had been a professional entity that could have helped them in their struggle with the educational system, they would have felt stronger.

Conclusion: It seems that cooperation between the families and professional entities could significantly improve their FQOL.

Advocacy for children with TS necessitates the combined work of a number of professional bodies and the family, and the parents envisaged that such assistance would significantly help families lower their level of stress and responsibility (Wang et al., 2004).

In addition, despite the parents' difficult daily coping with the various demands of caring for a child with TS, and despite the many concerns that they voiced, some parents indicated that the experience of raising a child with TS caused them to become better, more sensitive, helpful, supportive, open people and a close-knit family. The child with TS changed their life perspective:

He caused me to see myself as a good and understanding individual. I am much more patient with the other family members and very active in an organisation that helps other families.

The child with TS gave them strength to help other families with a child with TS and this gave them satisfaction. This finding is supported by the research of Li-Tsang et al., (2001), who found that families of children with disabilities felt that they had been blessed by the birth of their special child. They said that they had become closer and had become more compassionate, tolerant, sympathetic, flexible and selfless.

In some cases, the child with TS helped create a positive family interaction and a feeling of family unity. The child's very existence contributed to and helped family members better understand one another, and helped siblings be more tolerant and understanding:

When we discovered that our child likes to be out in nature, we began to plan hikes every week. The little children had a good time and he was calm and happy. This brought the family closer together and allowed quality time with the children.

Conclusion: the child with TS may constitute a source of family harmony and closeness, if the family members increase their acceptance and understanding toward each other and siblings are given appropriate information so that they can become more involved.

6.1.5 The severity of the syndrome emerged as a critical FQOL component for families who have a child with TS

The research found that all the families feared uncontrolled outbursts by the child with TS. The severity of the TS had a significant effect on the FQOL especially when the child with TS had 'strange' behaviour that embarrassed the parents. Parents reported that because of the severe behaviour of the child with TS, the family could not function as it had done before the appearance of the symptoms and preferred to remain isolated.

In addition the symptoms of the TS engendered much family tension which affected the quality of family relations: *'The many pressures we faced made our family life impossible.'*

To the extent that the child had what was perceived as a more severe case of TS and difficult and more violent behaviour, the stress that the family encountered was more intense. Parents noted that the situation sometimes led them to give up and abandon their attempts to deal with the situation:

I feel that I am not strong and am unable to help my children, and sometimes my response to the situation is introversion and silence.

The child's strange behaviour apparently embarrassed not only the family, but also the people around them:

He would jump and spin and you couldn't take him to the store. He would spin around all the way to the store and back home. He also had a tic that was like a dog barking and would attack. His behaviour was very severe, disruptive in school. The neighbours spoke about it. It also bothered the family. It was very annoying.

Additionally, the severity of the TS led the family to stop visiting friends and extended family:

The feeling that the environment blames me for not knowing how to raise my child and that I am not a successful mother is very difficult, in particular, when it comes from the people who are closest to you.'

And they preferred to stay at home in a safe place.

Because of the severe behaviour, parents needed a better diagnosis from good professionals which was usually very expensive:

At age 4 we went to a psychiatrist because he had temper tantrums, he would yell, he didn't stop repeating words over and over. The doctors diagnosed ADHD and recommended Ritalin.

Ritalin usually worsened the situation and the tics increased. Often it was only after years of suffering, tension and stress that the child was diagnosed as a child with TS.

The severity of the TS influenced the ability of parents to devote time to their other children and siblings felt neglected:

Attention and energy are too focused on the child with TS, and I feel that I am not sensitive to the other children. I feel that this is not fair, but I do nothing about it.

When a parent could not function because of the severe behaviour of the child with TS, the family experienced much stress:

The stress of our lives tore my husband and me apart, not an uncommon story in the life of a child with TS. We could not understand and we needed guidance to go through all this. We couldn't function as a family.

In some cases when one parent could not bear the child's behaviour and decided to abandon attempts to cope and to leave, the responsibility for the family fell on the remaining spouse who underwent a lot of pressure:

My husband said he could not stand it anymore, and he said me or the child. So I said to him, you need a vacation. Take your things and go to Jerusalem, and then he left and came back a few months later.

The severity of the TS also had an impact on the child's status in the educational framework. If the situation was more difficult and more severe the school was more likely to be unable and unwilling to deal with the situation and the result is that the child is excluded from the system:

In Year 1 he didn't fit in, left the room yelled and cursed without end, spoke and spoke, didn't let the teacher teach. They asked us to find a different school for him and explained that it was not fitting for him to remain in the school.'

Conclusion: The findings indicated that for all the variables and among all types of respondents, assessment of FQOL was lower among families in which the child has a more severe case of TS and this was especially so in cases where the child with TS has more co-morbid disorders (Wilkinson et al., 2001). The assessment of services for the family and the child with TS was low especially the 'support and counselling' and 'information' variables. These findings are supported by Wilkinson et al., (2001), who indicated that TS symptom severity was significantly correlated with the level of impact on the family and with the number of co-morbid disorders. They also indicated that TS is a disorder with effects that extend beyond motor and vocal tics.

CHAPTER 7: CONCLUSIONS

The goal of this study was to investigate the FQOL of families who have a child with TS and to understand how the existence of a child with TS living at home affects the FQOL. Therefore, the main questions addressed by this study were:

1. Which FQOL domains are perceived by family members to be affected by the fact that they are raising a child with TS?
2. How do different families raising a child with TS perceive their FQOL?

7.1 Factual Conclusions

The evidence collected for this research answered the research questions and identified what appear to be five new sub-domains: ***Advocacy, Cooperation, Explanation, Information*** and ***Severity of TS*** that have a significant impact on the FQOL of families who have children with TS.

The integration of the findings that emerged from the various research tools, and in reference to the five QOL sub-domains (Turnbull et al., 2004) seem to allow the advancement of the five identified sub-domains as sub-domains of FQOL, a finding unique to this study. These variables are essentially thematic categories that emerged as a result of the interpretation and the discussion of the findings.

Figure 4 below presents these thematic categories.

Figure 4: The ACEIST Model: New FQOL Sub-domains involved in the FQOL of Families who have a Child with TS

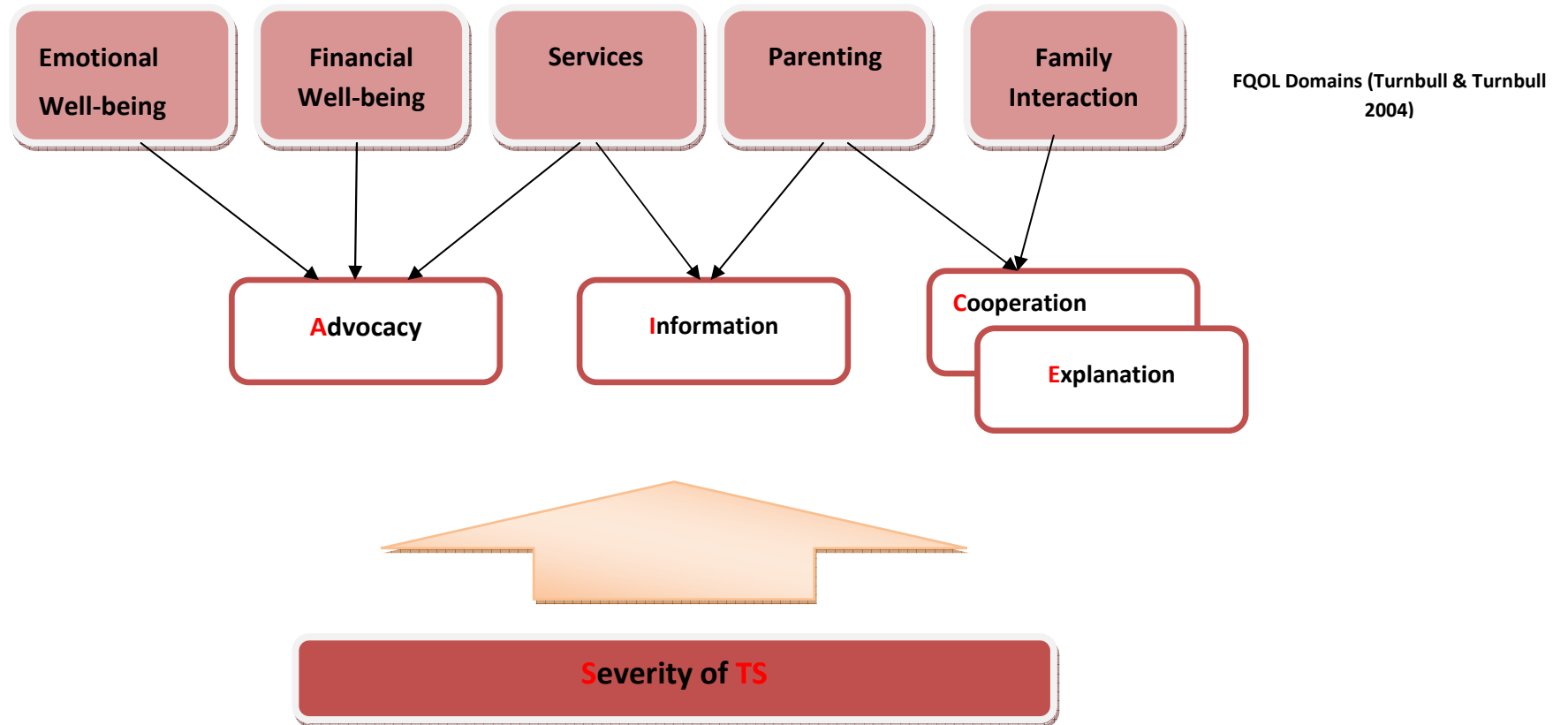


Figure 4 shows the **ACIEST** model, including the five newly identified sub-domains emerging from the research findings, namely, **Advocacy**, **Cooperation**, **Information**, **Explanation**, and **Severity of TS**.

'**Advocacy**' emerged in both the 'emotional well-being' and 'services for the child and family' domains of FQOL. This sub-domain relates to the need to advocate for the child with TS in relation to the society. In particular, it relates to the need to explain to and influence educational professionals, extended family and friends in order to provide the best possible care and protection for the child with TS. '**Cooperation**' emerged in both the QOL domains: 'family interaction' and 'parenting'. Cooperation relates to the ability of the parents to cooperate in order to fulfil their parenting role by creating a warm and positive atmosphere in the home and by preventing feelings of tension and stress, and siblings' feelings of neglect and isolation. '**Information**' emerged in both the QOL domains: 'parenting' and 'services for the child and family'. This sub-domain relates to the parents' need to obtain true and correct information from doctors and counsellors who know and understand the disorder. It seems that accurate information could prevent inappropriate behaviours toward the child with TS and siblings and assuage parents' guilt feelings so that parents could act and perceive themselves as good parents. '**Explanation**' emerged in both QOL domains: 'Family interaction' and 'Parenting'. 'Explanation' relates to the process in which parents struggle to explain the phenomenon of TS to their other children about TS.

According to the research findings, 'The **Severity** of the **TS**' seems to constitute a variable that amplifies the level of difficulty of each of the newly identified sub-domains. Thus, for example, as shown previously, if the severity of the TS is greater, there will be a greater need for Advocacy, in order to maintain the FQOL of families who have a child with TS.

7.2 Conceptual Conclusions: The ACEIST Model

On a conceptual level, the research advances the ACEIST model as a new way to view the FQOL of families who have a child with TS, providing new understanding concerning the components of their FQOL. This model presents an accurate and applicable approach to this issue that can be employed as a tool in order to assess and examine the FQOL of these families through research.

The research seems to indicate that the main need of these families is to receive true, accurate and comprehensive **information** from professionals concerning the nature of the TS and its implications. Accurate information about the syndrome at the early stages of diagnosis can apparently prevent parents from acting inappropriately or even violently towards their children with TS in order to 'discipline' them, and allow better understanding of the child's situation. Parents blamed themselves for many years for their violent behaviour towards the child with TS, and felt that they were 'not good' parents. However, when parents received accurate information about the syndrome, it helped them to organise their priorities correctly, and understand that what was really important was to provide the help that the child with TS needs. Additionally, having accurate information apparently helped the parents to stand up to their extended family and friends with more power when they needed to deal with prejudices regarding their child with TS. If explanations did not help and grandparents and aunts and uncles did not change their attitude concerning the behaviour of the child with TS, the parents at least felt more comfortable after they knew the facts about the syndrome and what should be done to help the child, even if it meant distancing themselves from extended family and friends.

Once they had accurate information about the syndrome, most parents seemed able to maintain a balance in the family and could provide **explanation** and support to the other children. Parents felt that they would also be able to plan their child's treatment at the early stages of the emergence of the symptoms if they had the proper information.

Knowledge of the symptoms of the syndrome at an earlier stage could guide the parents' reactions towards their children, and if this were successful they would be able to perceive themselves as successful parents, and help them serve as a better and more positive example to their other children.

As it was, the parents often lacked sufficient information and support so that it was found that parents felt that they neglected their other children and were unable to fix this. They were often not sensitive enough towards the other siblings, and did not share their emotions or information with them. If they were given accurate information, they seemed to be more able to explain the syndrome to their other children and thus prevent misunderstandings, and consequent unpleasant situations and confusion among their children in all areas relating to the syndrome. Siblings would not have to speculate regarding the nature of the problem their brother or sister had. They would not feel so ashamed when facing their friends, and they would not unnecessarily fear that the difficulties of their sibling with TS indicated that the child was going to die. Transmission of accurate information to siblings would open channels for family communication and **cooperation** and thus decrease unnecessary stress.

It appeared that all families benefitted from coordination which enabled them to maintain a 'normal' life. However, when it comes to families who have a child with TS, **cooperation** between the parents seems to be critical. A child with TS needs much attention, as well as medical and psychiatric treatments. Most of the time, he needs to be accompanied at school and outside the house. Parents of children with TS face many challenges in raising a child with TS. Therefore, **cooperation** between them could decrease stress and tension, employing open conversations that could prevent feelings of isolation. Cooperation between the parents meant that there was an effective division of roles between them and that the burden did not fall only on one of the parents. Such a relationship apparently sustained parents and enabled them to deal with the day-to-day stresses providing tools to decrease tension. It seems that when there is adequate cooperation, parents do not feel so exhausted and have more time and energy to give attention to the other children in the family, and siblings will not feel neglected.

In addition, it seems to be essential for parents to have accurate information about the syndrome so that they can act as **advocates** for their children when facing the teachers and other professionals who work in the educational framework and have relations with their child with TS. In order that these professionals can help the child with TS to integrate and function within the education system, they need to be sensitive to the difficulties of the child and those of the parents. The findings of the present research show that in most cases the professional functionaries of the education system give up and send the child with TS home, leaving him without any educational solution. Advocating for their child with TS, it appears that the parents must be equipped with relevant knowledge so that professionals and the decision makers in the educational framework can also understand the problem better and be more willing to deal with the difficulties. In this case, it would be easier if the professional bodies involved could see the child with TS as a child who needs emotional, behavioural and sometimes academic support.

In the present research, the educational framework was found to be the most influential variable affecting FQOL for families of children with TS. Parents expressed their dissatisfaction with the educational framework, but also expressed hope that if the educational system could learn to deal with the child with TS, then there would be an improvement in their QOL.

7.3 Recommendations

The conclusions that emerged from this study seem to allow the advancement of several recommendations that can serve as a framework OR model to help families, educators and therapists cope more effectively with the difficulties of a child with TS. Lack of accurate diagnosis and information may lead families to a state of imbalance and great stress. Professionals should understand that their role is very critical to these families. Inaccurate or lack of diagnosis leaves families with a feeling of helplessness when dealing with the misunderstood behaviour of their child with TS. The result is a state of stress that permeates to each family member, and thus negatively impacts the family as a unit.

Additionally, the educational framework seems to avoid and reject any attempt to understand the important role it plays in the lives of these families. In the Israeli reality children with TS are not defined as children special needs. This research indicates that children diagnosed as having TS need to be included within this special group in order to receive appropriate educational and therapeutic assistance. Inclusion within this definition would help to improve the FQOL of those children's families. This research offers this knowledge as a recommendation for government education policy in Israel.

The results that emerged from this research also have implications for the policy of the Ministry of Health. It is recommended that family doctors and paediatricians receive training to raise their awareness and knowledge regarding children with TS in order to avoid parents' exhausting searches for accurate diagnosis and advice and medical treatment with consequent saving of family's financial resources. Evidently, the need to constantly supervise the child with TS in order to prevent unsafe behaviours meant that parents often neglected the child's siblings. Families ceased their activities outside the home because they did not want to hear the comments of neighbours and friends. It is recommended that awareness towards TS should be raised amongst the community through the social services in order to avoid ignorance regarding this syndrome. Advocacy should not be the exclusive realm of the parents, but should also be employed by professionals, who can explain the syndrome and assist the education system in their work with the children with TS.

7.4 Suggestions for Further Research

It is recommended that a thorough research should be conducted using the new ACEIST model of the FQOL of families who have a child with TS.

The research findings indicated that information and explanations concerning TS would have helped siblings in their daily coping within the family and with their friends in school. It is implied that there is a need for additional in-depth investigation of siblings' perceptions regarding the FQOL of families with a child with TS since to the best of the researcher's knowledge no previous research has focused on the experiences of siblings of children with TS.

Additionally, more in-depth study is needed to examine, on the one hand, how parents of children with TS perceive the educational framework, and on the other hand, to examine teachers' perceptions regarding coping strategies that could be employed with children with TS in school. Such a study could perhaps provide possible answers to the difficulties involved in the integration of children with TS in the education system, and form a foundation for an intervention programme in the educational framework which could help parents of children with TS.

Future study could try to replicate this study using a larger sample to provide more specific and richer information concerning the perceptions of families raising a child with TS of their FQOL.

It is important to examine the impact of the severity of the TS on perceptions of FQOL in a more profound/ OR extensive research.

It is also considered important to make a comparison between the perceptions of the child with TS regarding his own FQOL and perceptions of other family members in order to obtain a multi-faceted understanding of this issue.

7.5 Contribution of this Research to the Corpus of Knowledge

This section indicates the contribution of the research to knowledge. It returns to the various aspects of the gap in knowledge that were described at the beginning of the thesis, and shows how the research embodies novel and original conclusions and theory, and the relevance of these conclusions for governmental policies for education, social action and health in Israel.

7.5.1 Contribution to Extant Knowledge

This research has answered the research questions and filled the gap that was identified in knowledge by providing factual evidence and conceptual conclusions concerning the ways in which FQOL of families who have a child with TS is affected by the existence of this child in the family.

The gap in knowledge was filled through an examination of the families' perceptions of their FQOL, and the influence on this FQOL due to the existence of a child with TS in the family. The research and its conclusions drew on knowledge concerning behavioural psychology (Barlow et al., 2001), systems theory (Bowen, 1978; Birnbaum et al., 1997) and the ecological model (Bronfenbrenner, 1979; Oetzel et al., 2006) which provided theoretical underpinnings for the study.

The discovery of what appears to be new influential sub-domains of QOL that affect the FQOL, created a new understanding that enabled the researcher to construct the ACEIST model, and suggest new theoretical ideas concerning this issue.

By filling the gap in knowledge the research contributes new knowledge concerning FQOL, providing original insights upon this issue, relevant for Israeli policy-makers at a national level, and their plans for educational, social, and health policies.

The new ACEIST model represents a novel and original view of FQOL of families having a child with TS. This new model may have practical implications for the design of training frameworks educational, social, and health staffs to create new ways of assisting these children and their families.

Since no other research was found that combined the theories from the different fields of knowledge: behavioural psychology, systems theory and the ecological model within an integrated conceptual framework (Trafford, 2007), to examine the FQOL of families who have a child with TS; this too constitutes an original contribution of the present research.

Epilogue, My Ph.D. Journey

When I decided to continue with my Ph.D. studies, I thought that this would involve many hours of independent study and the reading of voluminous materials and many hours of summarising, slowly gathered enough material to write my dissertation. But it was not so.

The process of choosing a topic for my research was not difficult, since a number of years beforehand, when I had previously considered continuing on with my doctoral studies, I knew that if I decided to continue, the subject would relate to children with TS. At first I wanted to examine the QOL of children with TS. A change in direction of the research towards FQOL among children with TS occurred as a result of the fact that I wanted my research to provide a true and in-depth picture of FQOL of families of children with TS, so that I could construct an intervention programme that would help them in the relevant areas, based on their own views. Therefore, the research was constructed on a more systemic outlook, including the entire nuclear family, parents and siblings.

Thus began my search for research tools dealing with family systems. I began the process in 2005, and discovered during my search for materials and research dealing with FQOL, that until the beginning of the 2000s, researchers had not conducted research on FQOL in the context of children with various types of disabilities. I was astounded to discover that the first study that examined FQOL among children with mental disabilities was published in 2004. With mixed feelings, I understood that while my decision to study family systems was correct, this appeared to be a new field and researchers had possibly avoided studying families because of the problematic and complex nature of the topic. However, I also understood that if I was truly interested in this issue, I would rise to the challenge. It was preferable to do something new, challenging and interesting, rather than doing 'just another doctorate.' From the moment that I made the decision regarding my research topic, my journey began.

The research significantly improved my level of thinking on a number of planes. I learned how to ask the correct, appropriate and most contributing/ OR rewarding/ OR profitable questions. Correct questions helped me not only in my field of study but also in my daily life and at work. I discovered that many things depend on asking the correct questions. I discovered that asking the correct questions contributes to improved interpersonal interaction, family life, and work. (That could be an interesting subject for research).

Conducting the research taught me to use organisational strategies. I learned to organise material and to use my time efficiently. This was also true on an occupational level. The process of gathering information from interviews, organising material into tables by categories, and summarising the material (which was a long and arduous process), gave me ideas for improving the organisation of material in my own work, and helped me develop a new learning strategy that I have been passing on to teachers and students whom I teach.

The research improved my written English and generally improved my level of writing, which is now more understandable, flowing, and considerate of the reader. I learnt that nothing can be taken for granted, and it is necessary to write clearly and in an understandable fashion so that other people who are not knowledgeable in the field of education or psychology could understand my writing.

On a personal level, today I can say that my research helped me to become a more sensitive, open, understanding and accepting person. The research, and in particular the interviews, which brought me in contact with families in their homes, showed me aspects of family life that I did not realise were so strong and important. I discovered that families have great power and the ability to cope with any situation. The power of words articulated in conversations and interviews cannot be transmitted to the reader of the research. It was difficult to transmit body language, tears and silences in the room, looks that passed between parents, tone of speech and the occasional breakdown.

At the beginning of my research I was concerned that the interviews would not be deep enough and would not provide a true picture. I was worried that families would not want to cooperate and would not speak. I was worried that they would talk about concrete matters that could not contribute to my research. I was astounded by the force of information that the families provided. I was astounded that it was almost unnecessary for me to intervene in the conversation. I asked the question: 'Tell me what it is like to live with a child with TS in the home' and they began to speak and did not stop. They spoke about themselves, about the siblings, about the extended family and about the educational framework. They moved from topic to topic without stopping. There was a feeling as if they had been waiting for someone to come and ask them questions so they could continue to speak. Their need to speak and to tell was so strong that sometimes I felt as though I should stop them, because on one hand I did not know how to cope with all the information, the emotions and the power that I was receiving, and on the other hand, I was concerned I would not be able to cope with all the data later.

Going into homes, entering families gave me mixed feelings of happiness and sadness. I was happy that I could bring back much research material and achieve deep insights in order to explain what really affected the families. I was sad because it was difficult to see how some of these families suffered, but still continued to cope. Sometimes, at the end of an interview, I would leave the home of a given family, stand by my car, shocked, with tears in my eyes, take a deep breath, and continue on.

Today, after completing my doctoral dissertation, I am pleased with my decision to continue with my studies and the decision I made in choosing this topic, and I hope that I shall be able to construct an intervention programme to assist these families in their daily coping. I hope that my work will contribute to an increased awareness to the needs of families who have a child with TS among various professionals, and in particular within the educational framework, which does not understand the important and dominant role it plays in the quality of life of families who have children with TS.

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APPENDICES

APPENDIX 1: THE FAMILY QUALITY OF LIFE SURVEY

The questionnaire that appears here was developed by the Beach Center on Disability, at the University of Kansas, in partnership with families, service providers and researchers and adapted specifically for the research population described in this thesis: i.e. families who have a child with TS.

Survey Information and Instructions

Thank you for agreeing to complete this survey.

This survey has questions about:

- the services you and your child need and/or receive,
- the things that make your life together as a family good,
- you and your family in general

We will use your answers to help us improve policies and services for children with TS and their families.

All the information you give us is confidential. Your name will not be attached to any of the information you give us. It is important that you answer as many questions as you can, but please feel free to skip those questions that make you feel uncomfortable.

When answering these questions, please think about your experiences **over the last 6 months**.

Thank you so much for sharing your opinion with us!

By completing this survey, you indicate that you have been informed of the important aspects of this study and you are willing to participate.

Part One: Support and Services for THE CHILD WITH TS

1. Where does your child usually receive services? (Mark only one circle)

- ☐ At home
- ☐ At a community preschool or childcare centre
- ☐ Public school
- ☐ Private School
- ☐ Special or alternative school
- ☐ Other community setting
- ☐ Work setting
- ☐ Other _____

2. The degree of necessity and services received for the child with TS

Which of the following services does your child currently need ?			If YES , what level of service does he or she actually receive ?		
	Yes	No	None	Some	Enough
Special equipment to help your child learn, and grow	o	o	o	o	o
Health services (medical evaluations, nutrition, nursing)	o	o	o	o	o
Hearing and/or vision services	o	o	o	o	o
Physical and/or occupational therapy	o	o	o	o	o
Speech and/or language services	o	o	o	o	o
Special education services	o	o	o	o	o
Counselling and psychological services	o	o	o	o	o
Behaviour support	o	o	o	o	o
Transportation and/or mobility services	o	o	o	o	o
Self-care skills	o	o	o	o	o
Other (please describe _____)	o	o	o	o	o

Part Two: Services for your FAMILY:

Which of the following services does your family currently need ?			If YES : what level of service does your family actually receive ?		
	Yes	No	Enough	Some	None
Respite care	0	0	0	0	0
Child care	0	0	0	0	0
Money to help pay bills	0	0	0	0	0
Homemaker and/or housekeeping services	0	0	0	0	0
Transportation services	0	0	0	0	0
Support groups	0	0	0	0	0
Counselling	0	0	0	0	0
Sibling support	0	0	0	0	0
Parent or family training	0	0	0	0	0
Information about specific disabilities	0	0	0	0	0
Information about where to get services for your child	0	0	0	0	0
Information about legal rights	0	0	0	0	0
Other (please describe) _____	0	0	0	0	0

Part Three: Family Quality of Life

In this section of the survey, we want you to tell us how you feel about your life together as a family. We will use what we learn from families to improve policies and services for children with disabilities and their families. Your 'family' may include mother, father and siblings.

Step 1: Importance – First, please shade in the circle in the first set of columns to show how important you think that statement is.

- Shading the **first** circle means you think the statement is only **insignificantly important**.
- Shading the **last** circle means you think that statement is **critically important**.

Step 2: Satisfaction – Please shade in the circle in the next set of columns to show how satisfied you are with that statement

- Shading the **first** circle means you are **very dissatisfied**
- Shading the **last** circle means you are **extremely satisfied**.

Please remember to grade both IMPORTANCE and SATISFACTION for each question.

Thank you so much for sharing your opinion with us!

Family Quality of Life Questionnaire

For my family to have a good life together...	This matter/ OR need has the following level of importance					How satisfied am I that this need is fulfilled?				
	Insignificant	Low importance	Important	Very Important	Critically Important	Very dissatisfied	Dissatisfied	Satisfied	Very satisfied	Extremely satisfied
My family enjoys spending time together.	0	0	0	0	0	0	0	0	0	0
My family members help their children to learn how to be independent.	0	0	0	0	0	0	0	0	0	0
My family has the support we need to relieve stress.	0	0	0	0	0	0	0	0	0	0
My family members have friends or others who provide support	0	0	0	0	0	0	0	0	0	0
My family members help their children with schoolwork and activities	0	0	0	0	0	0	0	0	0	0
My family members have transportation to get to the places they need to be.	0	0	0	0	0	0	0	0	0	0
My family members talk openly with each other.	0	0	0	0	0	0	0	0	0	0

My family members teach their children how to get along with others.	0	0	0	0	0	0	0	0	0	0
My family members have some time to pursue their own interests	0	0	0	0	0	0	0	0	0	0
My family solves problems together.	0	0	0	0	0	0	0	0	0	0
My family members support each other to accomplish goals.	0	0	0	0	0	0	0	0	0	0
My family members show that they love and care for each other.	0	0	0	0	0	0	0	0	0	0
My family has outside help available to us to take care of special needs of all our family members.	0	0	0	0	0	0	0	0	0	0
Adults in my family teach the children to make good decisions.	0	0	0	0	0	0	0	0	0	0
My family gets medical care when needed.	0	0	0	0	0	0	0	0	0	0
My family has a way to take care of our expenses.	0	0	0	0	0	0	0	0	0	0
Adults in my family know other people in the children's lives (friends, teachers, etc.).	0	0	0	0	0	0	0	0	0	0

My family is able to handle life's ups and downs.	0	0	0	0	0	0	0	0	0	0
Adults in my family have time to take care of the individual needs of every child.	0	0	0	0	0	0	0	0	0	0
My family gets dental care when needed.	0	0	0	0	0	0	0	0	0	0
My family feels safe at home, work, school, and in our neighbourhood	0	0	0	0	0	0	0	0	0	0

Please answer the following questions about your child with TS. If you have more than one child with a disability, please keep in mind the one who has the most impact on your FQOL.

For my family to have a good life together...	This matter/ OR need has the following level of importance					How satisfied am I that this need is fulfilled?				
	Insignificant	Low Importance	Important	Very importance	Critically Important	Very dissatisfied	Dissatisfied	Satisfied	Very satisfied	Extremely satisfied
My family member with TS has support to accomplish goals at school	0	0	0	0	0	0	0	0	0	0
My family member with TS has support to accomplish goals at home.	0	0	0	0	0	0	0	0	0	0
My family member with TS has support to make friends.	0	0	0	0	0	0	0	0	0	0
My family has good relationships with the service providers who provide services and support to our family member with TS	0	0	0	0	0	0	0	0	0	0

Part Four: General Individual and Family Information

The last thing we need to do is to ask a few questions about you and your family. We will use this information to generally describe the people who responded to our survey. We will describe people in groups, never as individuals, so your answers will be kept confidential. Please answer these questions about **yourself**.

1. What is your gender?

- ☐ Male
- ☐ Female

2. In which year were you born?

3. What is your employment status?

- ☐ Working- full time
- ☐ Working part-time
- ☐ Unemployed but looking for work
- ☐ Not employed (staying at home, parent or care giver, retired, disability, public assistance pay).

4. What is the highest level of education that you have completed?

- ☐ No schooling completed
- ☐ Formal schooling but no high school diploma
- ☐ High school diploma
- ☐ College diploma
- ☐ B.A diploma

- ☐ M.A or more

General information about the child with TS

5. What is your relationship to the child with TS in your family?

- ☐ Parent (Biological or Adoptive)
- ☐ Sibling

6. What is the gender of your child with TS?

- ☐ Male
- ☐ Female

7. In which year was your child with TS born?

8. What is the degree of severity of your child's TS?

- ☐ Mild
- ☐ Moderate
- ☐ Severe

9. What is the nature of your child's **secondary** disability?

- ☐ ADD or ADHD
- ☐ OCD
- ☐ Emotional or behavioural disorder
- ☐ Hearing impairment including deafness
- ☐ Learning disability

- ☐ Mental disability
- ☐ Speech or language impairment
- ☐ Vision impairment
- ☐ Health impairment (Please specify) _____
- ☐ Other disability (Please specify) _____
- ☐ No specific diagnostics

General Family Information

According to the Central Bureau of Statistics, the average income per family in Israel is 8,500 NIS.

10. What was your total household income from all sources for the past year? Be sure to include income from all sources (such as family subsidy or child support).

- ☐ Much more than average
- ☐ More than average
- ☐ Average
- ☐ Lower than average
- ☐ Much lower than average

11. How many people are supported on this income?

- ☐ 1 ☐ 5
- ☐ 2 ☐ 6
- ☐ 3 ☐ 7 or more
- ☐ 4

Thank you! You have finished completing this survey. Please make sure you erase any extra marks and that you have answered all the questions.

APPENDIX 2:

REQUEST FOR PERMISSION TO USE THE BEACH CENTER RESEARCH, EVALUATION, AND PLANNING TOOLS

Date: August 3, 2005

Name of tool(s) you are requesting permission to use: FQOL Survey

Your Name: Rita Khoury

Organisation: Anglia Ruskin University, UK

Address: Zipory St. 37 Nazareth Elite, Israel 17502

Phone: 00-972-0524-466339

Email: hrita@netvision.net.il

In order to ensure that our research, evaluation and planning tools are used for the purpose they were intended and with the underlying values clearly stated, we ask that you provide a brief description of your intended use for the tool(s) named above.

What is the overall purpose of your research or evaluation?

I would like to evaluate the QOL of families of children with TS. This research is for my PhD.

Please briefly describe the target population, sample size, and general characteristics of the sample (age, type of disability, location).

The questionnaire will be distributed among all the families who have a child with TS in Israel and who agreed to participate in the research (about 25-30 families). The questionnaire will be given to the parents of a child with TS and to the two siblings that are close to the TS child's age.

What date do you want to start collecting data?

In October, 2006

When do you anticipate completing data analysis?

Between October and December 2007

Are you willing to providing the Beach Center with an electronic copy of the data from this instrument as well as the demographics from the sample? Yes

Any alterations or changes to the tools must be agreed to by the Beach Center. Occasionally such changes are necessary to reach an audience slightly different from that on which the tool was developed, and we are quite willing to cooperate with you in determining acceptable changes. Please describe any changes you would like to make to the tool. I would not make any changes in this tool. Just translate it to Hebrew.

If you have any questions concerning this request, please contact Denise Poston at the Beach Center at 785-864-7601 or denisep@ku.edu.

Please submit the completed request to Denise Poston at the Beach Center by mail, fax or email.

Beach Center Tool Kit

Attn: Dr. Denise Poston

3111 Haworth Hall

1200 Sunnyside Ave

University of Kansas

Lawrence, KS 66045

Fax – 785-864-7605

Email to denisep@ku.edu

APPENDIX 3: PARTICIPANT INFORMATION SHEET

Purpose and value of the study

The quality of life of families who have children with TS has not been studied yet in Israel. Although it is known from studies about TS Syndrome, that these children suffer from serious difficulty in their daily adjustment and coping within the framework of the family, a lack of awareness among professionals who are directly or indirectly in contact with the child and his family also reflects on the family's ability to cope.

Such studies as this one can raise awareness of the issue, and can provide the opportunity to develop an intervention programme that will facilitate improvement in families' quality of life.

Invitation to participate

You are invited to participate in semi-structured interviews at a location of your choice.

Who is organising the study?

The sole researcher for this study is Rita Khoury

What will happen to the results of the study?

The results gathered at the interview will be transcribed and analysed for use in the above-mentioned student's PhD studies dissertation. Thereafter, the material collected by the researcher will either be destroyed or archived according to your wishes.

Source of funding for the research

The researcher will be funding this project.

Contact for further information

You can contact the researcher to discuss any matters regarding the project and the interview according to the following contact addresses:

Home Tel: 04-6465635 Mobile: 0524-466339 Email: hrita@netvision.net.il

Section B: Your Participation in the Research Project

Why you have been invited to take part

The sample group needed for the interview and to complete the questionnaires was identified by the researcher as families who have a child with TS.

Whether you can refuse to take part

You can refuse to take part at anytime

Whether you can withdraw at any time and how

You can withdraw at any time by simply completing the withdrawal slip at the bottom of the consent form and forwarding it to the researcher.

What will happen if you agree to take part?

If you would like to be involved in this research project and agree to complete a questionnaire and to be interviewed, please coordinate a suitable time and place when you are free with the researcher. The interview should take no more than two hours. The interview will be tape-recorded unless you have an objection to this. Please inform the researcher of your preference regarding the recording. If at any time you feel uncomfortable with the questions being asked please inform the researcher. Once the interview has come to an end the researcher will ask if there was any material that you would prefer to be withdrawn from use in the research project. If so, this will be noted and the material will be discarded as soon as possible.

Are there any risks involved (e.g. side effects from taking some substance) and if so what will be done to insure your wellbeing/safety

Due the sensitive nature of the research, the interview will cover personal information regarding your family and your child. If at any point during the interview you feel uncomfortable regarding the subject matter please inform the researcher and the interview will stop immediately.

Agreement to participate in this research will not compromise your legal rights should something go wrong

There are no special precautions you must take before, during or after taking part in the study.

What will happen to any information/data/samples that are collected from you?

Any material collected by the researcher will be analysed and used in the in the main body of the dissertation and thereafter will be destroyed or archived in accordance with your wishes.

Are there any benefits from taking part?

The benefit from your participation in this study would be your contribution to understanding and facilitating family coping with a child with TS, an area that needs more research and study.

In addition, your involvement with this project will help the researcher in achieving a PhD certificate.

How will your participation in the project be kept confidential?

Your participation in the project will be kept strictly confidential. These are the ways in which the researcher intends to do this:

Anonymity At the beginning of the interview the researcher will offer you a pseudonym to be used instead of your actual name. This will then be used throughout the interview and throughout the entire research project.

Confidentiality Before the interview the researcher will discuss issues of confidentiality. First, you will be informed how the information will be kept confidential. The researcher will explain that all tapes, transcripts, notes and any other material taken during the interview will be secured in a locked filing cabinet. The information collected during the interview will be available to you when you request. The researcher will also discuss with you what will happen to the data collected from the interview, after the project has finished. The researcher will discuss the possibility of keeping the data for future research. If you have any objections to this then the data will be destroyed, including all tapes, notes and descriptions.

All the above will be recorded on an ethic consideration sheet and attached to your notes and transcripts.

YOU WILL BE GIVEN A COPY OF THIS TO KEEP, TOGETHER WITH A COPY OF YOUR CONSENT FORM.

APPENDIX 4: PARTICIPANT CONSENT FORM

NAME OF PARTICIPANT:

**Title of the project: Perceptions of FQOL of families who have a child with
Tourette syndrome**

Main investigator and contact details:

Rita Khoury

Mobile: 052-4466339

Email: hrita@netvision.net.il

Members of the research team: Just me, Rita

1. I agree to take part in the above research. I have read the Participant Information Sheet which is attached to this form. I understand what my role will be in this research, and all my questions have been answered to my satisfaction.
2. I understand that I am free to withdraw from the research at any time, for any reason and without prejudice.
3. I have been informed that the confidentiality of the information I provide will be safeguarded.
4. I am free to ask any questions at any time before and during the study.
5. I have been provided with a copy of this form and the Participant Information Sheet.

Data Protection: I agree to the University¹ processing personal data which I have supplied. I agree to the processing of such data for any purposes connected with the Research Project as outlined to me*

¹ "The University" includes ARU and its partner colleges

***Note to researchers:** please amend or add to this clause as necessary to ensure that it conforms to the relevant data protection legislation in your country

Name of participant (print).....Signed.....Date.....

Name of witness (print).....Signed.....Date.....

YOU WILL BE GIVEN A COPY OF THIS FORM TO KEEP

If you wish to withdraw from the research, please complete the form below and return to the main investigator named above.

Title of Project: Perceptions of FQOL of families who have a child with Tourette Syndrome

APPENDIX 5:

EXTRACTING KEY PHRASES FROM THE TRANSCRIPTS

Interview with Family Number 3 - 25.1.07

Family from Haifa. Two parents and three children: an eight-year-old boy with Tourette, and a six-year-old girl, and a four-year-old boy who do not have Tourette. The younger boy has begun blinking, and all the children have been diagnosed with ADHD. The mother has ADHD and dyslexia and also face twitches. She may have Tourette. The father has been diagnosed with Tourette with ADHD.

The mother works at home as an interior designer. The father owns his own business.

Rita: What is it like living with a child with Tourette?

The mother spoke without my stopping her and said the following:

G. was a difficult baby, crying all the time, always nursing, not sleeping, and constantly moving. He wore me out and I did not understand why it was happening. I was a young mother and I didn't know if this was normal or not. As a baby, he didn't sleep, moved all the time, jumped from place to place, and I couldn't leave him for a minute because I was afraid he'd fall.

At age 4 we went for a psychiatric consultation, because he had temper tantrums, he would yell, he didn't stop repeating words over and over. The doctors diagnosed ADD and recommended Ritalin.

The severe problem began in school, in Year 1 he didn't fit in, left the classroom, yelled and cursed without end, spoke and spoke, didn't let the teacher teach. They asked us to find a different school and said that it did not suit them for him to stay in school. We found an open school. After we found an open school and he was accepted, he cursed a child and called him 'dirty Ethiopian'. The child was the son of the therapist in the school, and they called and said they could not keep him because of his cursing [with tears in her eyes]. We didn't know what to

do. I knew that I had to fight for him. I asked the Tourette Organization to come to the school and lecture the teachers on the phenomenon. I called the Educational Support Centre and asked for an aide to accompany him. At that time he had many temper tantrums and yelled and cursed and spoke all the time. He kept repeating the word "sex". In every sentence and every situation, he would say "sex".

One day his younger sister was punished at school and they moved her into G.'s class. G. stood up and said "My sister has sex with my little brother all the time. My sister has sex ... has sex..." and she started to cry and felt horrible.

He does not have any learning disabilities. When he is calm he is smart and fits in well. Because of his attention disorder and hyperactivity, he can't study. In the first year, the teaching assistant was not good and did not help. G. did not like him. In the second year, the relationship was better. Every time the teacher couldn't cope with him, she would ask the assistant to take him out into the schoolyard. At home he would go crazy and would not stop yelling and cursing. He would say: B. S. is a rapist, rapist, rape, rape," all the time, without stopping. Sometimes he would be quiet, but sometimes he would drive me crazy and I would put him in a room and ask him not to come out.

We went to a psychiatrist and she gave him Halidol, Concerta and Ritalin. There is an improvement in his attention, but he still curses and has tantrums.

I am really afraid that the day will come when I won't be able to grab him and calm him down.

I am particularly afraid of adolescence.

He will grow up, continue cursing, and I won't be able to control him. ... it really scares me. Now he is small, but what will happen when he grows up (tears in her eyes, choking slightly while speaking)? Sometimes I think that he will become a criminal, and I don't want to think about it.

If I hadn't turned to those responsible in the Ministry of Education and the Educational Support Centre, the school would have kicked him out, and he would be in special education, a school for disturbed children. It would have destroyed him, because he's a smart boy. At

the age of one year and four months, he was speaking in full sentences. He has no learning disability. What luck! He has no problems with obsessive behaviour.

I work here at home because I have to be available. They call from school all the time. Sometimes I have to go and get him and bring him home. In Year 1, we spent most of our time in school, my husband and I. It was really difficult. Now, with the aide, things are calmer.

Our mornings are a nightmare. He doesn't move, does not do anything, doesn't get dressed, and does not eat anything. He's like a VIP. You have to dress him and he doesn't cooperate. He's like an unmoveable rock, going crazy and cursing. In the morning he wakes up without the influence of any medication and he doesn't function. He just yells and curses. We only give him his medication after breakfast do and the influence of the medications begins after he leaves the house. On the way out of the house, on the way downstairs, on the stairway, he yells, and the building shakes from his yelling. At night he goes to sleep late and it's difficult for him to fall asleep.

Externally, he's a good looking and a charming child. He has no motor tics. So until he starts to curse, he looks like any other normal boy. People don't understand that he has a disorder and look at him strangely. They think he's not disciplined. I don't have the strength to explain it to everyone.

The recommendation of the Tourette Organization was that I let him be and that I should not continually tell him to stop, because he doesn't control his behaviour. But the psychologist said we have to set boundaries and teach him that it's not nice and not okay, and with time, he'll learn to control himself. Now he is more in control of himself in school, but at home he has outbreaks, curses, and doesn't stop moving. Only the computer calms him down. It worries me that he spends hours sitting in front of the computer, but it seems to calm him down.

Rita: Is there anything else you want to share?

Mother: At first we had some difficult times. We didn't know what to do, and it created tension between us. But now, I feel that it's brought us closer. My husband has Tourette. We discovered this only after we understood that G. suffered from Tourette. We speak a lot. The only problem is financial. As a young couple, it was hard financially, and all the treatments and the psychologists cost us money. We also paid for the teaching assistant, because very little came from the Educational Support Centre. Now we have more money and I bring in a nanny, and she helps. I can then continue on with my day. In the evening I get a babysitter and we go out to a movie or a restaurant. That's okay. So that we can have enough money, my husband works longer hours, from eight to eight. Twice a week I go out to an exercise and yoga class and my husband puts the children to bed. That also helps. I am satisfied with myself, and that is important to me.

The mother continues: I think that I am a good mother. I invest and I love. I have difficult days. I give punishments. Once I gave more punishments. Today I am more understanding.

The only help we get from the family is financial. My mother and father visit rarely. They say that I don't know how to discipline my children. My husband's mother is a very old woman and does not really interfere. We have enough financial help.

All our friends know. There are those who had difficulty with the cursing and stopped coming over. One friend keeps blaming me saying that I give my son medications and medications aren't good. What she doesn't understand is that I have no choice. She would make me upset with her comments and I told her to stop coming by. She made me feel guilty. What kind of friend is that? But the rest are okay. We don't have enough time for friends.

We go out to a movie or a restaurant together, and it's fun.

On Saturdays we go out hiking into nature and G. really likes it.

The Researcher's Personal impressions: The family has serious difficulty in daily functioning, expressed particularly in the morning. As the mother said, "mornings are a nightmare". There is noise, and a lack of order, G. doesn't function in the morning at all, he has to be dressed, fed and dragged to school. It is very difficult and does not allow normal functioning. The mother is under stress when she thinks about what could occur or happen during the day when he's at school. Her tone was one of desperation, even though she tried to explain that she was functioning. The house is a mess, there is no order. Papers are scattered on the floor. I didn't see any food in the kitchen even although I was there during the afternoon hours. Most of the work is done by the mother. She's exhausted. There is almost no furniture.

A most obvious and painful need was evident at school. The educational system did not know how and did not want to cope with the problem. They threatened the mother and referred her to all sorts of frameworks which were not suitable for her child. She was miserable and did not know what to do or who to turn to. When she talked about that period, there were tears in her eyes.

According to her, money is the solution. She pays a teaching assistant in school, brings a nanny to help at home with the children in the afternoon and a babysitter when she goes out with her husband. If they didn't have money, it could be much worse.

Words that the mother used: frustration, helplessness, misery, lack of function, nightmare, fear of the future and the present.